The British Journal of Surgery

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THE
BRITISH JOURNAL OF SURGERY

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CARCINOMA OF THE FLOOR OF THE MOUTH*

BY SIR STANFORD CADE, K.B.E., C.B.
WESTMINSTER HOSPITAL, LONDON

CARCINOMA of the floor of the mouth is a clinical entity only in its early stages. Involvement of adjacent structures is common and soon the disease spreads beyond the floor itself. Its mode of spread can be conveniently but somewhat arbitrarily considered as in three directions—to the intrinsic and extrinsic muscles of the tongue, to the gum with or without involvement of the alveolus, and to the cervical lymph-glands. Although such a subdivision is artificial it is of help in considering the choice of treatment and in the planning thereof (*Fig.* 267).

The tumour itself follows the usual course of epithelial cancer in the mouth; it is histologically an epithelioma varying in degree of keratinization and anaplasia; it is either an exuberant mass or an ulcer with indurated edges. The disease is most common in men (about 80 to 85 per cent). Although dental sepsis is found in a number of patients, many tumours develop in edentulous mouths. The usual so-called precancerous changes such as leucoplakia, chiefly of the floor itself and of the gingival mucosa, are frequently seen. All these features are well known and require no emphasis.

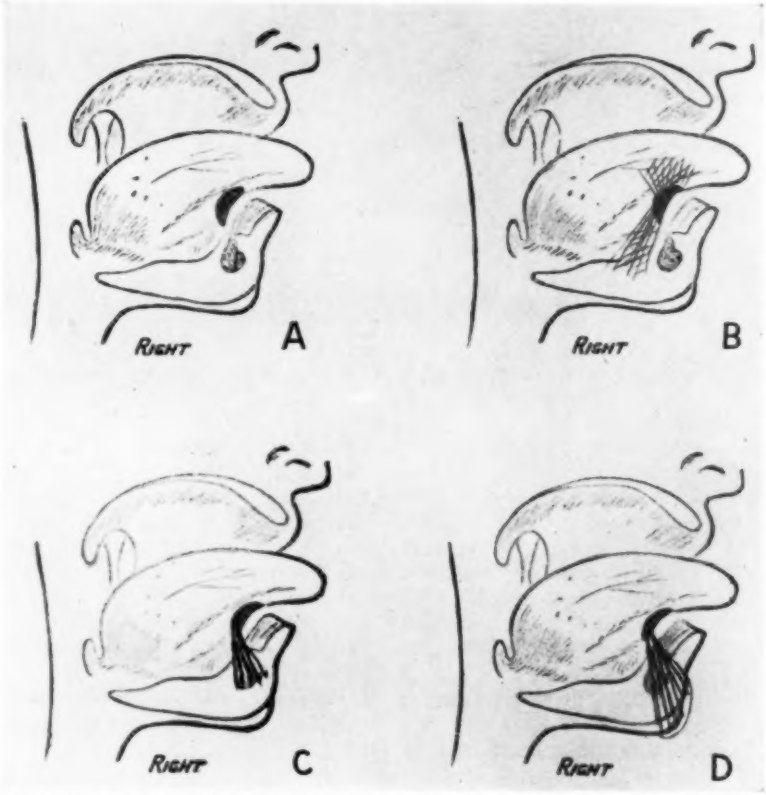

FIG. 267.—Diagram indicating extent of disease. A, Floor of mouth only; suitable for radiotherapy. B, Extension tointrinsic and extrinsic muscles of the tongue; suitable for radiotherapy followed by diathermy. C, Extension to mandible; suitable for surgery without bone-grafting. D, Extension through jaw to skin; suitable for excision and grafting.

* Read before the Association of Surgeons, Cardiff, April 19, 1952.

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DIFFERENTIAL DIAGNOSIS

It is necessary to rule out chronic inflammatory conditions, salivary calculi, specific infections, and this differential diagnosis follows on the same lines as in tumours in other sites in the mouth. But in addition attention should be drawn to four other conditions which may cause some difficulty in diagnosis in some patients: (1) *Benign mucous-and-salivary gland tumours* primarily situated in the floor of the mouth; these may involve the mucous membrane and cause ulceration from pressure

A B

FIG. 268.—Lesions of floor of mouth involving gum. A, Metastasis from a primary carcinoma in the tongue. B, Primary carcinoma of the floor of the mouth.

atrophy; (2) *Acute monocytic leukæmia* with ulceration in the mouth and enlargement of the cervical lymph-nodes; (3) *Metastasis* in the lower jaw and adjoining gum and mouth floor from a clinically silent primary growth in the tongue (*Fig.* 268); (4) A primary malignant growth in the mouth may be a *sarcoma* of the rhabdomyosarcoma type. All these four conditions can mimic the more common epithelial cancer. If X-ray examination of the jaw and of the chest, a full blood-count, and a biopsy are carried out as a routine the diagnosis can be established with accuracy.

A further point of interest in the natural history of cancer of the mouth floor is the occurrence of a second and independent primary growth, sometimes many years after the appearance of the original lesion in the mouth. In the series of cases under review such tumours have occurred in the urinary bladder, rectum, bronchus, œsophagus, or elsewhere in the mouth and pharynx, sometimes many years after successful treatment of the growth in the floor of the mouth.

FAMILIAL INCIDENCE

Familial incidence has occurred in two sets of brothers and one set of sisters, who presented themselves either simultaneously or at short intervals with similar or identical lesions in the floor of the mouth (*Figs.* 269, 270). *Figs.* 269 and 270 illustrate two sisters who came to the Westminster Hospital on the same day with identical histories of soreness on the left side of the mouth of a few months' duration. Both had ulcerating lesions on the left side of the mouth floor involving the alveolar mucosa and both had metastases in the left submandibular region.

Two brothers developed a carcinoma of the right side of the floor of the mouth extending posteriorly towards the pharynx. There was two years' interval between the onset of the disease; both were in the late sixties and the elder brother survived the younger one by two years.

Twin brothers developed lesions in the floor of the mouth involving the tongue at three years' interval. Both were chronic alcoholics, both subsequently developed metastases in the cervical lymph-nodes. One twin later developed a phalangeal metastasis in the big toe and the other a separate and probably independent œsophageal cancer. All three sets of brothers and sisters died of the disease.

MATERIAL

During the period 1925–51 there have been in all 118 patients with lesions in the situation under consideration. Of these, 97 patients seen up to 1947 lend themselves to a 5-year period of observation. A study of these patients may be of help in assessing the efficacy of the methods of treatment employed and in pointing out where changes in treatment technique are indicated. This quarter of a century covers the period of active and rapid development of radiotherapy and equally a period of considerable progress in surgical technical advances, made possible by the modern methods

of anæsthesia, antibiotics, and the technique of reconstructive or plastic surgery. Looking back at all this it cannot be denied that some progress has been made, but cancer of the floor of the mouth still remains a formidable problem and no single

METHODS OF TREATMENT

The methods of treatment available are radiotherapy or excision. The author's experience of radiotherapy is nearly entirely by radium needling or teleradium; neither intra-oral radium applicators

FIG. 269.—Two sisters (not twins) who developed simultaneously a carcinoma on the left side of the floor of the mouth with metastases in the left submandibular region.

FIG. 270.—Carcinoma of the floor of the mouth in the two sisters illustrated in *Fig.* 269.

method can claim pre-eminence in its control. One observation is, however, important: half-hearted attempts by any method are doomed to fail; inadequate surgery or radiotherapy by a single piece of apparatus or a single technique will hardly ever lead to control of the disease.

nor X rays at the conventional 250 kv. or the low-voltage contact X rays have been extensively used nor have they given sufficiently encouraging results to warrant a change from radium or surgery.

Early Localized Lesions.—Radiotherapy is the method of choice in the early localized lesions. In

such cases it can be curative with greater ease, less mutilation, and less risk than by surgical excision. When the disease is limited to the mucous membrane of the floor of the mouth or to the submucosal areolar tissue, interstitial radium alone is sufficient. For many years radium needles or radon seeds have been used; a more recent technical advance is the use of platinum shielded radio-active tantalum wire; this new development enables an easier and more accurate distribution of the radio-active source in the confined space of the floor, especially in its lateral extensions; it also permits simultaneous interstitial radiation of the submandibular and submental areas and the intra-oral lesion with great accuracy.

FIG. 271.—Deformity following wide excision of floor of mouth in advanced lesions. The whole lower lip, the skin, the chin, and the jaw have been removed. A tube-pedicle has been raised on the left shoulder. This method of delayed repair is badly tolerated by cancer patients.

Localized Spread to Muscles.—Where the disease has spread to the surrounding muscles either into the inferior surface of the tongue or to the extrinsic muscles of the floor, radium either interstitially or externally by teleradium, should be followed by local diathermy destruction of tissue at an interval of four to eight weeks after radiation. The tissue dose should be no less than 7000 r. At times 9000 r or even 10,000 r can be given, although with such doses the incidence of delayed localized radio-necrosis of the mandible is greater; such risk is justified and worth while if control of the disease is thereby achieved. The immediate radium burn from gross overdosage must be guarded against by careful technique.

Extension to Alveolar Mucosa.—When the lesion has extended to the gum mucosa, but the bone is clinically and radiologically intact, teleradium should be used in preference to needles; if available supervoltage X rays at 2 M.E.V. or more is equally efficacious. Complete regression of the tumour and sound healing can be achieved.

Radio-resistance.—Failure to control the disease, that is primary failure, when lack of satisfactory response is noted after adequate radiotherapy, occurs in a percentage of cases. It occurs more frequently in the keratinizing and less malignant tumours than in the anaplastic embryonic type. Rhabdomyosarcoma is also completely radio-resistant. It is important to recognize this type of case early, face the situation, refrain from submitting the patient to repeated radiotherapy, and consider surgical excision as soon as the lack of response to radiation is noted.

Involvement of Mandible.—Bone invasion by an epithelial cancer of the floor of the mouth is an indication for surgical treatment as the first choice. It is known that some patients with bone involvement do respond to radiation and patients considered inoperable or unsuitable for surgery should be so treated. Nevertheless, surgery is considered the method of choice. Patients in whom surgical excision is contemplated fall into two groups: The first group, where the growth is still limited in extent, permits of preservation of the skin and subcutaneous tissue and therefore immediate closure of the wound without any grafting procedure. Access to the lesion must be wide; this can be achieved by a reflection of a suitable whole-thickness flap consisting of the lip, chin, and part of the cheek; removal of the bone must be adequate; the submandibular triangle should be cleared of lymph-glands and of the submaxillary salivary glands, even when these are not obviously involved. Closure should be in two layers: (1) Accurate

approximation of the mucous membrane reinforced by the submucosal tissue and muscle between the cut cheek and the tongue; this should be watertight; (2) Closure of the skin with drainage. The deformity resulting from the removal of half the lower jaw is surprisingly small. Bone-grafting is not a routine procedure even when the central portion of the lower jaw is removed. In cases of cancer of the floor of the mouth bone-grafting should be delayed till primary healing of the soft tissues is achieved and the scar is stable. Bone-grafting in such cases should be done as an independent and separate procedure when freedom from recurrence is likely and not before an interval of six months following excision. In such patients a suitable lower denture often obviates the need for a bone-graft.

The second group consist of patients where the disease has involved the lower jaw extensively, including the central portion, where the alveolar mucosa of the gum, the sulcus between this and the lip, and subcutaneous tissues are involved. Here very wide excision with sacrifice of the lip and skin is essential (*Fig.* 271). The deformity resulting from such operations is very great. Patients with cancer, the elderly age group, tolerate prolonged multiple-stage plastic operations very badly. Attempts at an immediate repair in such cases necessitates some sacrifice of the cosmetic result, but the earliest possible closure should be aimed at. The preliminary raising of tubed pedicles from the shoulder or chest (*Fig.* 271) is not recommended. Even in the hands of the expert plastic surgeon this method needs many operational sessions, takes several weeks if not months, and the survival of the flaps is uncertain. The repair should be done by bringing a forehead flap down, lining it with skin from the neck and Thiersch-grafting the resulting raw surface (*Fig.* 272). Co-operation between the surgeon who carries out the ablation of the cancer and the plastic surgeon is an absolute essential.

Lymph-node Involvement.—Lymph-node involvement is dealt with on the same principles as in cancer of the tongue. If the primary growth is controlled, the lymph-nodes enlarged but operable, a block dissection is carried out. If the lesion is in the midline or mainly unilateral, although transgressing the midline, the necessity for a bilateral block may arise. The side most heavily involved is operated on first. An interval of three weeks should be allowed before the second side is operated on. Although block dissection is indicated it should only be done if the result of treatment of the primary intra-oral lesion is satisfactory and likely to remain so. When the glands are adherent or fixed, the operation is very unlikely to control the spread and radiotherapy to the lymph-node area should be used, mostly as a palliative. Teleradium alone or combined with needling is a satisfactory method and is preferable to X-ray therapy at 200 or 250 kv. Supervoltage X-ray therapy is more effective than the lower voltage and in some patients as effective as teleradium.

Inoperability.—In view of the magnitude of the surgical procedure cases submitted to surgery should be of reasonably good prognosis. Criteria of inoperability should be kept in mind. The following are considered inoperable: (1) When the disease has spread posteriorly and involved the pharyngeal mucosa or the pharyngeal part of the tongue; (2) When the lymph-nodes are adherent or fixed

FIG. 272.—Extensive removal of entire lower jaw and chin, lower lip, and part of the upper lip in advanced carcinoma of the floor of the mouth. Immediate repair by turning down a forehead flap and lining it by skin from the neck. Further operations are needed to complete closure. Compare with *Fig.* 271. (The repair in these two patients was done by Mr. J. M. Reidy.)

to the main vascular sheath; (3) When there is œdema of the subcutaneous tissue; (4) When there is gross involvement of the soft tissues in the submandibular or submental areas (*Fig.* 273). In such cases palliative radiotherapy has a great deal to offer and local moderate diathermy can keep the disease under control for a time.

RESULTS

Of the total of 97 patients treated by all methods 8 patients were lost sight of in less than 5 years. This leaves 89 cases suitable for assessment, and of those 24 remained well for periods of 5 or more years, that is 27 per cent. *Table I* shows the subdivision of cases.

Table I.—CARCINOMA OF THE FLOOR OF THE MOUTH. SURVIVAL

Extent	*Total Number of Cases*	*5-year Survival Number*	*Percentage*
Floor only	22	10	45
Floor and tongue ..	23	4	17
Floor and jaw	12	5	41
Floor and lymph-nodes	32	5	15
Total	89	24	27

The table shows that in early and localized lesions a 5-year survival of 45 per cent has been achieved by radium therapy. When the mandible was involved surgical treatment with and without radiation has given 41 per cent of 5-year survival. Involvement of the tongue by direct extension or spread to the cervical lymph-nodes reduced the 5-year survival to 15 or 17 per cent.

CONCLUSIONS

Early and small localized growths should be treated by radiation only. Local involvement of tongue and gum requires local diathermy in addition. Involvement of bone and widespread disease in the mouth is offered a better chance by surgical excision. Surgery should be on a large scale. Repair should be well planned and, except for bone-grafting, the local closure should be accomplished as quickly as possible. Methods using tube-pedicles are badly tolerated by patients with mouth cancer, and other methods, such as rotation or sliding flaps, quicker in achievement although of lesser cosmetic effect, should be employed. Immediate closure is the aim and the closely planned co-operation with a plastic surgeon is of utmost importance. Such extensive, mutilating, and complicated surgical procedures are in essence teamwork, where each member of the team is of equal importance.

Progress in the future will depend: (1) On early diagnosis; (2) On technical, clinical, and biological advances in radiotherapy; (3) On surgical advances, especially in the methods of closure of wounds.

FIG. 273.—Carcinoma of floor of the mouth involving mandible and fungating through skin. There is lymphatic invasion of the skin well shown in the lateral view. Although technically excision can be carried out, such cases are considered inoperable as however wide the excision local recurrence soon follows operation.

CARCINOMA OF THE FLOOR OF THE MOUTH

By Professor B. W. WINDEYER, London

This contribution to a symposium on cancer of the floor of the mouth is based on a study of the patients who came for treatment to the Middlesex Hospital since 1931. With the cases of cancer of the floor of the mouth I have also included those which involved the lower alveolus, as in many instances it was impossible to determine whether the site of origin was floor of mouth or alveolus, and there seemed to be similar problems involved in their treatment. There were in all 131 patients who came to the hospital during the years 1931 to 1946, the latter being the last year on which 5-year statistics can be based.

AETIOLOGY

Age.—The age distribution was similar to that which has been described frequently in other forms of cancer of the mouth and throat. The youngest was 38 and the oldest was 82. The decade 60–69 accounted for almost one half of the total (*Fig.* 274).

Sex.—There were 118 men and 13 women.

Associated or Predisposing Conditions.—Probably the most frequent associated abnormality in the mouth is patchy leucoplakia both on the floor of mouth and alveolus, and also on the tongue and buccal surface of the cheeks. Many lesions appear to have begun in such a patch of leucoplakia. When teeth were present they were usually in very poor condition, with gross infection around the gums. It is difficult to say whether such dental sepsis had any particular association with the cancer or whether it was a usual finding in hospital patients of this age group during this time.

The presence of leucoplakia and of dental sepsis at the time when the patient came for advice were by no means constant. In some instances the patient had been edentulous for many years and a not infrequent history, particularly in those cases where the lesion started primarily on the alveolus, was that a sore place developed underneath the denture which had become rough or ill-fitting.

The Brown-Kelly or Plummer-Vinson syndrome with generalized atrophy of the intrabuccal and pharyngeal mucosa was present in several of the women patients.

The total of 131 cases consisted of :—

- 53 in which the primary lesion involved floor of mouth alone.
- 16 in which the primary lesion involved the lower alveolus alone.
- 62 in which the primary lesion involved the floor of mouth and lower alveolus.

There were 52 with no glandular metastases on clinical examination and 79 in which there were clinical glandular metastases.

The histological diagnosis is almost invariably squamous-cell carcinoma. The majority have well-marked keratinization. There were in this series 3 cases of squamous-cell carcinoma with no keratinization and 3 cases of undifferentiated carcinoma. In cases treated in later years there have been tumours of mucous gland origin and recently a patient with papillary adenocarcinoma of the thyroid presenting as a recurrent tumour in the floor of the mouth following excision of a lump from the neck.

SYMPTOMS

The symptoms of which patients complained showed considerable variation. Some complained of feeling a lump in the mouth or of a feeling of discomfort, particularly in association with dentures which may have become ill-fitting. In others there were no symptoms referable to the mouth, and the lesion in some was found on routine inspection by a dentist. The appearance of a lump in the neck was in several cases the first intimation of any abnormality. More than one elderly patient with extensive intra-oral growth was referred to his doctor by relatives because his speech had become indistinct.

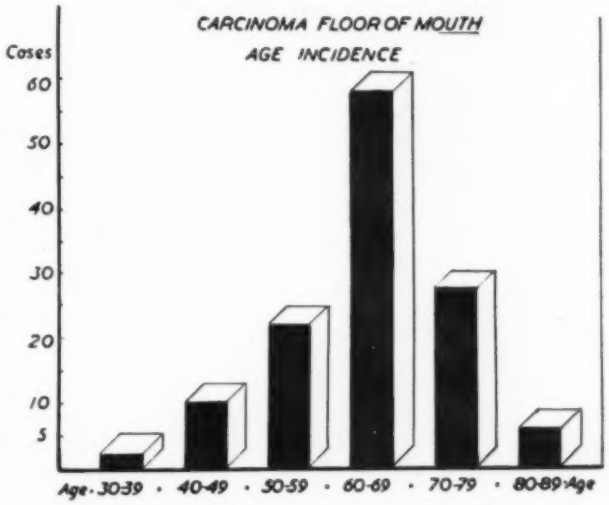


Fig. 274.—Age incidence of cancer of floor of mouth.

In some cases the lesion was exquisitely tender on palpation, but pain was not a marked feature in the case-histories unless the disease had become advanced with marked infection, breaking down glands, and extensive bone involvement. Toothache was complained of as the first symptom in several cases of alveolar involvement.

The most frequent early case is an ulcerated lesion with surrounding induration situated near the midline in the floor of the mouth, perhaps involving the frænum or associated with the orifice of one of Wharton's ducts (*Fig.* 275). It may spread forwards or laterally to be attached to or involve the mandible (*Fig.* 276) and a high proportion spread back to involve the deeper part of the tongue (*Fig.* 277).

The growths beginning on the alveolus may be ulcerating in type and extend within a short time

into the bone as well as to the floor of mouth and out towards the cheek (*Fig.* 278). Hypertrophic lesions may be found in both floor of mouth and alveolus (*Fig.* 279). They may be small and apparently quite superficial, but nevertheless may give rise to extensive glandular metastases (*Fig.* 280). It is in this type that the primary lesion is most likely to be overlooked and in which glandular metastases may be the first presenting symptom.

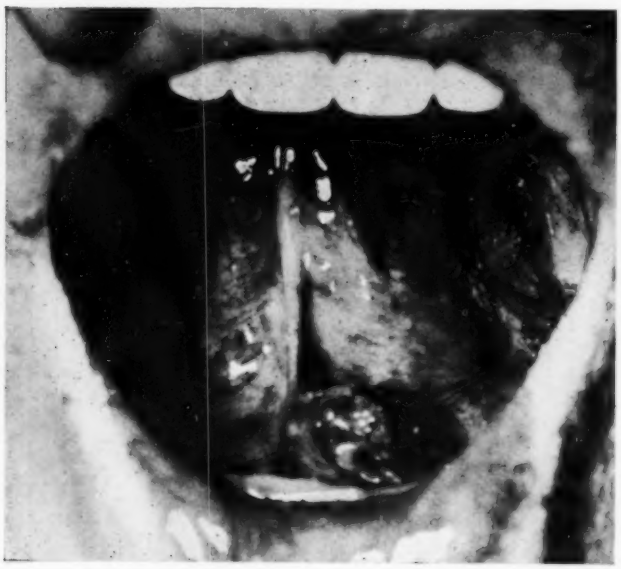


FIG. 275.—Early growth of floor of mouth.

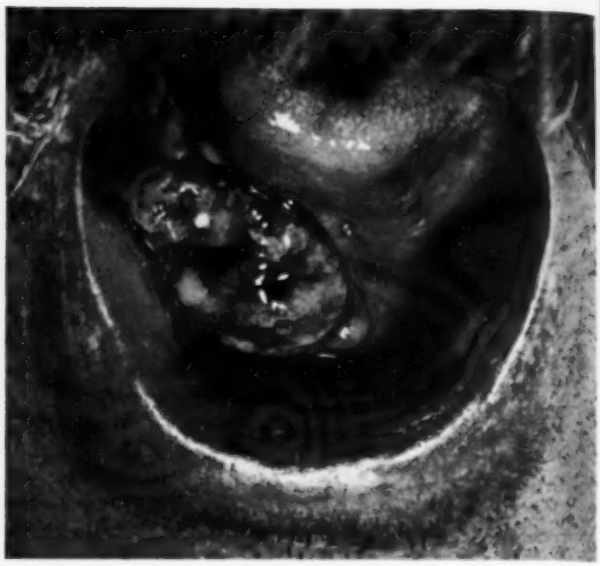


FIG. 276.—Growth of floor of mouth extending to mandible.

FIG. 277.—Growth of floor of mouth extending into tongue.

Nodular growths in the floor of the mouth, apparently arising beneath the mucous membrane and with extensive induration and early attachment to the jaw, are much rarer (*Fig.* 281). On account of the intact mucosa there may, in this variety, be greater delay in establishing the diagnosis.

In the later stages of the disease there may be spread outwards or downwards to involve and ulcerate the skin over the jaw or in the submental region. The ulceration within the mouth may become an extensive necrotic cavity, bounded by exposed mandible in front, by a fixed and indurated tongue behind, and filled by necrotic debris and food which, on account of its immobility and dependent position, remains a stagnant pool. The patient's general condition rapidly deteriorates and there may be continuous and intolerable pain. Glandular metastases may be unilateral or bilateral, involving submental and submaxillary groups or any part of the deep cervical chain. In this series approximately 60 per cent of cases had clinical glandular metastases.

FIG. 278.—Ulcerated growth of alveolus commencing on area of leucoplakia.

DIAGNOSIS

In the great majority of cases the diagnosis is straightforward and presents no difficulty, but there are certain cases in which an accurate diagnosis is difficult or impossible on clinical grounds and biopsy

FIG. 279.—Small hypertrophic growth of alveolus.

and histological examination is needed. Traumatic ulceration from a roughened tooth or badly fitting denture is the most frequent cause that I have seen for patients being sent up with a mis-diagnosis of cancer of the floor of mouth or alveolus. The patient shown in *Fig.* 282 had an ulcer which had been present for several months, caused by a sharp piece of metal protruding from a denture. It was hard and indurated but became soft and healed in three weeks when the denture was removed. Chronic trauma may cause a fibrous papillomatous mass (*Fig.* 283). It may resemble a hypertrophic cancer.

Such errors in diagnosis are not, however, of importance if care is taken to prove the diagnosis by biopsy, if necessary repeated biopsy, before major therapeutic measures are started. Of far greater importance were errors in the opposite direction. Several patients had treatment delayed until the growth had become advanced because cancer had not been thought of and no histological examination had been made. In several cases, after a diagnosis of traumatic ulceration, the dentures were left out, a period of observation ensued, and the growth became much worse. In one case the diagnosis was syphilis and because the Wassermann reaction was positive no biopsy was taken and a prolonged course of antisyphilitic treatment administered. In growths involving the alveolus, diagnosis of osteomyelitis following dental extraction and of actinomycosis have similarly caused delay. The value of biopsy is still continually being debated. It obviously has limitations, particularly when a negative result is obtained, but I am convinced that in this situation early biopsy is of the greatest importance in any suspicious ulcer or tumour.

TREATMENT

The total of 131 cases consisted of :—

53 in which the primary lesion involved floor of mouth alone, 26 without glands and 23 with glandular metastases.

FIG. 280.—Glandular metastases in same patient as in *Fig.* 279.

16 in which the primary lesion involved the lower alveolus alone, 11 without glands and 5 with glandular metastases.

62 in which the primary lesion involved both the floor of mouth and lower alveolus,

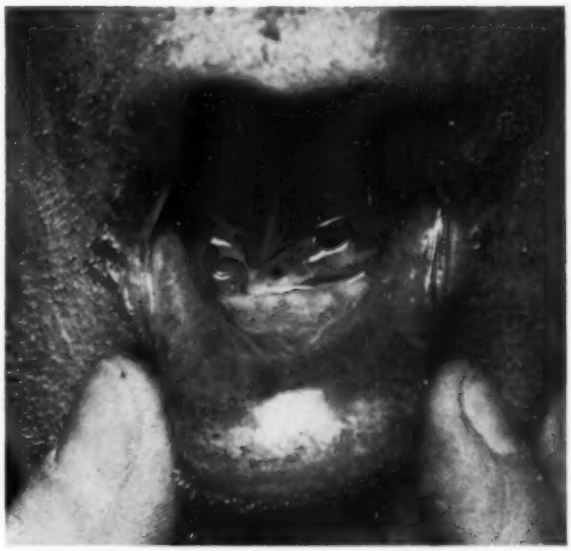


FIG. 281.—Nodular growth with fibrinous reaction from irradiation.

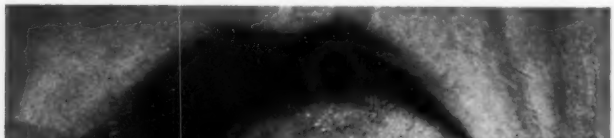
13 without glands and 37 with glandular metastases.

There were 52 cases with no glandular metastases on clinical examination and 79 in which there were clinical glandular metastases.

Thirteen cases were considered to be too advanced and were given no treatment; 3 were treated as regards the primary lesion by surgery; 115 were

treated as regards the primary lesion by various techniques of radiotherapy.

In the treatment of glandular metastases there was no completely consistent policy throughout these years. In general, when no glands were clinically



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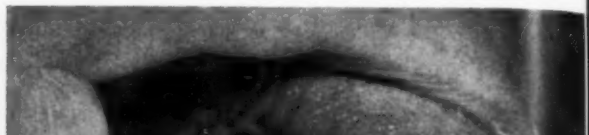


B

C

FIG. 282.—Traumatic ulceration from denture. A, On first examination; B, Three weeks after removal of denture; C, Denture with rough metal surface.

involved no specific treatment was given to the neck except such irradiation of the submental and submaxillary regions as occurred during the treatment of the primary lesion. When glands were palpable and metastases were considered to be present, block dissection was carried out in those cases which were considered operable and where the primary lesion was considered to have been controlled by irradiation. There were 14 patients who had block dissection. In more advanced cases radiotherapy alone was used to palliate and in some few cases to provide lasting regression.

Method of Radiotherapy.—Since 1932 up to the beginning of the war in 1939, and again during

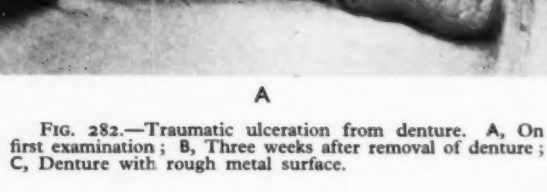


FIG. 283.—Fibrogranulomatous mass caused by irritation from a denture.

1945 and 1946, the main method of treating the primary lesion has been by a combination of intrabuccal radium applicator, teleradium, and, when necessary, a vertical single-plane radium implant across the mouth and posterior to the growth (*Fig.* 284).

The intrabuccal radium applicators which have been used were described by Greer Walker in 1938 and have not been greatly altered since then. They consist essentially of a moulded appliance which holds radium needles at a fixed distance, usually 7 mm., from the surface of the growth, irradiating the whole of the surface with, in addition, a margin of apparently healthy tissue. It is fixed firmly in position by dental appliances which vary according to the particular conditions. The tongue and remainder of the healthy tissues of the mouth are kept as far away as possible by the thickness of plastic material of which the applicator is constructed. We have used a lead box which is cast to enclose the needles. There has been argument as to the necessity and efficacy of this lead shield and admittedly it only cuts off about 10 per cent of the radiation, but I consider that even this is worth while as additional protection. By such an applicator the growth is irradiated from its superior surface.

The teleradium unit at present in use is one of 10 g. which has evolved in stages from a 1-g. unit in 1932 and a 4-g. unit in 1937. It is used to irradiate the lesion from below through the submental and submaxillary regions, and sometimes when the growth is spreading forwards or laterally additional fields may be directed horizontally to deal with spread to the lip or cheek. In cases with advanced glandular involvement, teleradium is also used to irradiate the neck.

The vertical implant has been placed through the anterior part of the tongue transversely across the mouth so as to irradiate any extension of the growth backwards into the deeper part of the tongue. Implantation of radium needles to the whole growth

was not used as the routine method of radiotherapy as we thought that it was more likely to produce necrosis of the mandible. Usually a tumour dose of about 7000 r is given by the combined treatment and it may be somewhat higher on the mucosa.

During the war years it was not possible to use the same techniques as radium was not available in London for some time, and the method of treatment was altered. X-ray therapy with a tension of 200 kv. and quality represented by 2 mm. Cu. half-value layer was used to irradiate externally in place of teleradium, and an intrabuccal X-ray applicator using a narrow beam and the same quality of radiation was used, where possible, to irradiate the primary lesion from above through the open mouth. Accuracy of application of this field was assured by the use of a periscope device which could be inserted through the wall of the applicator. Posterior extension of the growth was dealt with by an implant using radon needles. The doses used in a typical case were a minimum tumour dose of 6150 r, 3000 r being given through the open mouth and the remainder through the skin.

X-ray therapy and teleradium were used in some advanced cases as the sole methods of treatment to provide some degree of palliation.

In all cases it is essential to have the patient hospitalized as such patients are usually elderly and infirm, the treatment is prolonged over at least six weeks and is a considerable ordeal. They need skilled nursing, adequate rest, and special attention to their diet and fluid intake.

If teeth are present dental extractions are necessary, as I consider that the lower jaw should be cleared at the beginning of treatment. This may be necessary to obtain adequate application either of the intrabuccal radium mould or the intrabuccal X-ray applicator, but in addition it is necessary in order to eliminate infection and diminish the risk of subsequent necrosis. When teeth are present close to the area of ulceration a preliminary course of teleradium or X-ray therapy is usually given, up to about 1200–1500 r, before the extractions are carried out.

Of the 115 cases treated by radiotherapy to the primary lesion 33 were alive and free from recurrence at the end of 5 years, i.e., 28·7 per cent.

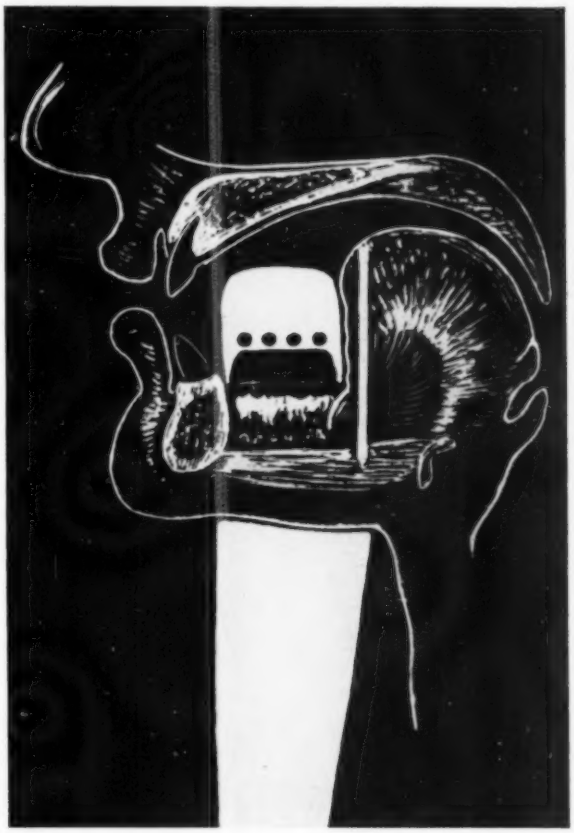


FIG. 284.—Diagrammatic representation of intrabuccal applicator, teleradium, and single-plane radium implant.

The crude rate of 5-year recurrence-free cases from all those coming to the hospital, including the untreated cases, is 35 out of 131, i.e., 26·7 per cent.

Analysis of the radiotherapy cases is as follows :—

RESULTS OF TREATMENT OF CARCINOMA OF FLOOR OF MOUTH

Total cases, including untreated	131	
Recurrence-free at end of 5 years	35	(26·7 per cent)
Treated by Surgery	3	
Recurrence-free 15 and 14 years	2	
Treated by Radiotherapy	115	
Recurrence-free at end of 5 years	33	(28·7 per cent)

Analysis of Radiotherapy Cases

	No glands		*Glands*		*Total*	
Floor of mouth :						
Cases	26		23		49	
5 years recurrence-free	14	(54 per cent)	4	(17 per cent)	18	(36·7 per cent)
Alveolus :						
Cases	11		5		16	
5 years recurrence-free	4		3		7	(44 per cent)
Floor of mouth and alveolus :						
Cases	13		37		50	
5 years recurrence-free	5	(38·4 per cent)	3	(8 per cent)	8	(16 per cent)
All sites :						
Cases	50		65		115	
5 years recurrence-free	23	(46 per cent)	10	(15·4 per cent)	33	(28·7 per cent)

RESULTS

Of the 3 cases treated by surgery, 2 remain alive and free from recurrence 15 and 14 years after the operation.

It is of interest to compare the results of the two main radiotherapy techniques :—

Radium		*X rays*	
Total	68	Total	47
5-yr. rec.-free	20 (29·4 per cent)	5-yr. rec.-free	13 (27·6 per cent)

I believe that the former technique is superior and I was surprised to find that in this series there is no appreciable difference in the results.

Length of Survival after Treatment (*Fig.* 285).—Of the total of 131 patients, which includes

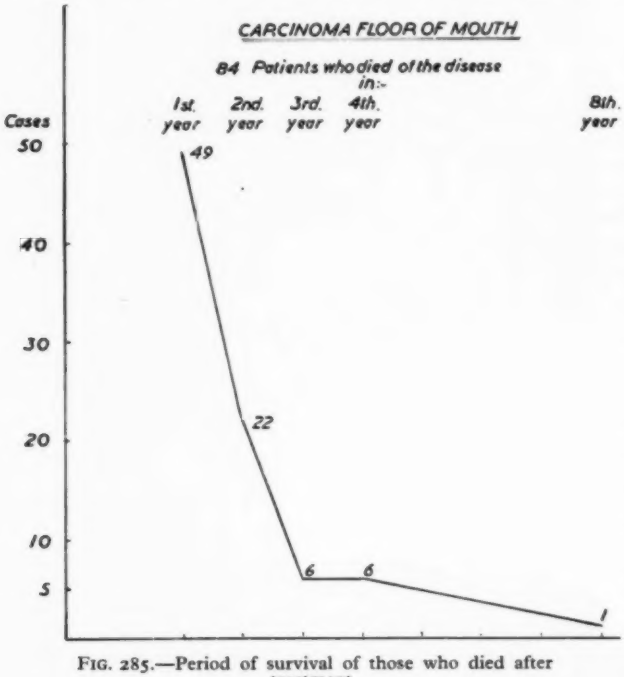


FIG. 285.—Period of survival of those who died after treatment.

the 13 patients untreated, 90 died of the evolution either of the primary growth or of glandular metastases. The exact date of death of 6 of them was not determined. Of the remaining 84 patients, 71 died during the first two years after treatment and the remainder before the end of the fourth year, with the exception of one who lived until the eighth year. Six patients died of intercurrent disease before the end of the fifth year and have been counted among the failures. Twelve patients developed at least one further primary cancer. In 6 of them it developed after five year's freedom from disease. There was 1 cancer of the bronchus, 2 œsophagus, 1 lymphosarcoma, 1 Hodgkin's disease. There were 7 patients with other primary lesions in the mouth. These may be debatable and may be considered by some to be recurrences, but I consider that they were more likely to be new primary lesions. One of them, aged 72 in 1938, had a keratinizing squamous-cell carcinoma involving the left side of the floor of mouth and alveolus treated by teleradium and implant. In 1940 he had a carcinoma of the middle third of the left side of tongue treated by radon implant and then partial glossectomy for localized necrosis. In 1946 he developed an anaplastic squamous-cell carcinoma of the epiglottis, from which he died.

Another patient whom I have considered as having developed a second cancer was admitted in 1931, for a carcinoma of the left side of the floor of mouth with palpable glands. He had a radium implantation to the mouth and a block dissection of the neck, the glands being invaded. In 1943 he developed a growth of the right side of floor of mouth which was treated by X rays and again a block dissection for cervical metastases. In 1951 he returned with a recurrence in the lower part of the neck. This was excised with a portion of the sternum. He has now developed a further recurrence which is receiving teleradium. The mouth remains free of growth although there had been a localized area of necrosis between the two areas irradiated (*Fig.* 286).

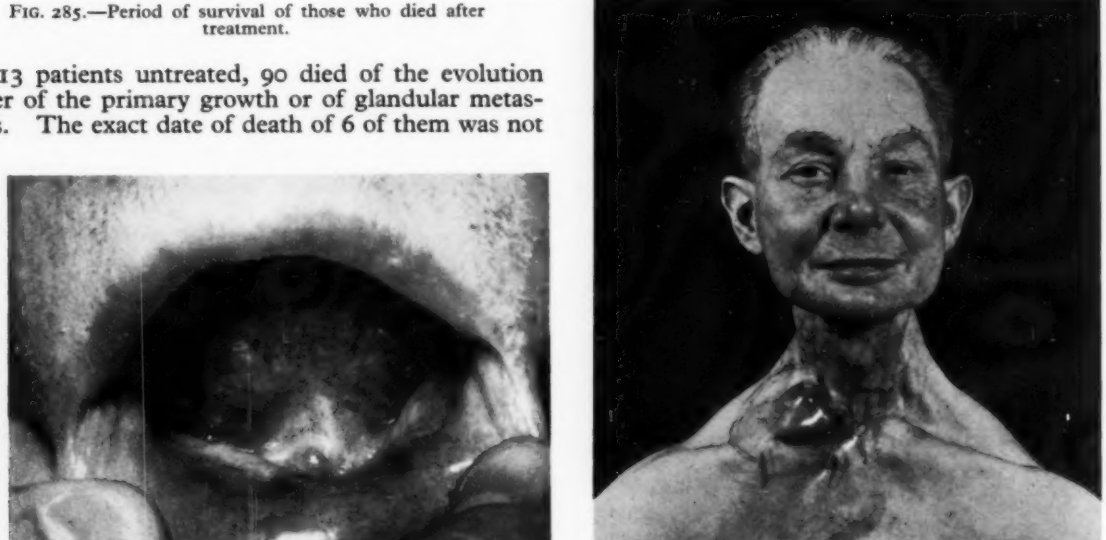
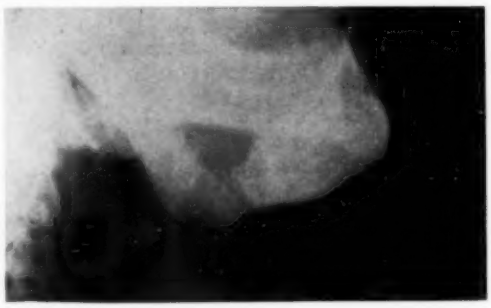


FIG. 286.—Patient who has survived 21 years. He has developed two growths in floor of mouth, has had bilateral block dissection, and now has recurrence in the neck.

COMPLICATIONS

Hæmorrhage from the ulcerated area is a complication which may occur during treatment in advanced ulcerated growths. It was the cause of death of one patient. Necrosis of soft tissues also occurred in the earlier years in some advanced cases due to infection and the difficulty of keeping clean the immobile ulcerated cavity. It does not present such a problem since the introduction of chemotherapy.



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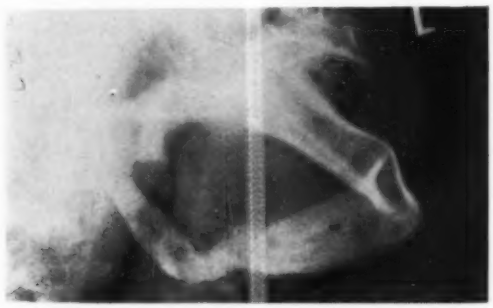

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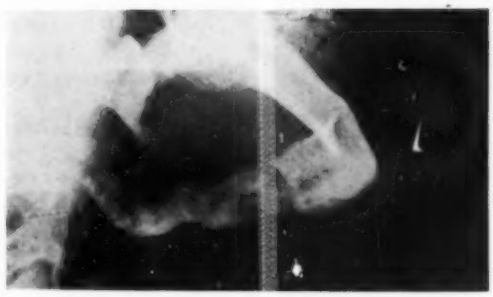

C

FIG. 287.—Carcinoma of the alveolus with involvement of mandible. A, Before X-ray treatment; B, Six months after treatment by X rays; C, Two years after treatment by X rays.

Necrosis of bone is the main complication of radiotherapy to be feared. It may occur soon after treatment or its development may be delayed even for years. It may be extensive and involve a large part of the mandible. It is usually accompanied by severe pain of long duration as sequestration is slow and any attempt at premature removal of the diseased bone may extend the process of necrosis.

There were in this series 11 patients who developed some degree of necrosis of either bone or soft tissue. In 3 of them it was severe and disabling over a long period. These three had all growths with radiological involvement of bone when first treated. They are all alive and free of recurrence more than ten years since treatment. In one of them, following sequestration of a portion of the inner surface of the mandible, there was little subsequent disability. The second developed a necrotic fistula into the submental region. It needs continual dressing and is a great nuisance, but he will not agree to any operation for repair. The third patient, a man aged 42, had an extensive keratinizing squamous-cell carcinoma involving the left side of the jaw with considerable infection and sinuses in the submaxillary and hyoid regions, at first diagnosed as osteomyelitis and then actinomycosis. He was treated entirely by X rays in 1941, as it was thought that surgical removal was impossible. A large portion of the left side of the jaw sequestrated and he developed an extensive fistula. Repair was delayed until 1944 when it seemed unlikely that there would be any local recurrence, and the defect was then closed with a pedicle graft. (*Fig.* 287.)

CONCLUSIONS

The analysis of this series of cases has not given an opportunity for discussion of the value of surgery in the treatment of the primary lesion in carcinoma of the floor of the mouth and lower alveolus. Two of the three patients treated by surgery had lasting freedom from recurrence, but the numbers are too small for any conclusions to be drawn. In the light of the experience gained by the treatment of these patients our present practice is to submit more patients to radical surgical excision. Cases of carcinoma of the alveolus with erosion of bone are most suitably treated in this way.

I consider that when bone is invaded surgical ablation should be undertaken whenever possible, even though it may mean an extensive procedure and the production of some deformity. Treatment by irradiation in such cases will always produce some degree of necrosis with its attendant pain and misery.

It is not, however, always impossible to destroy the cancer by irradiation when bone is involved and if excision is impracticable it may be well worth while, even though it means a prolonged period of pain and disability.

LEIOMYOMA OF THE ŒSOPHAGUS

By GEOFFREY FLAVELL, London

The rapid advance of thoracic surgery and the ever-widening use of mass radiography lend to the recognition and differential diagnosis of mediastinal tumours a lively interest and importance: for although many are long symptomless, it is precisely during the asymptomatic period they are most readily and safely removable; all are likely eventually to produce symptoms by secondary pressure effects upon the vulnerable structures around them; and 10 per cent of the whole are potentially malignant. Such tumours, rarely recorded in the not-far-distant past when the chest was surgically inviolate, or described only as freakish autopsy findings, now appear more and more often in surgical accounts, and it is clear that in another decade some radical revisions of our notions of their incidence will become necessary. In the meantime individual experiences accumulate, some of which may clarify diagnosis, modify misconceptions, and point to rational surgical treatment. Of these tumours œsophageal leiomyoma combines apparent rarity, equivocal symptoms, great diagnostic difficulties, and important surgical consequences.

Sporadic descriptions of these innocent œsophageal tumours have been cropping up in medical literature ever since 1559 when Sussius recorded one filling the lower third of the gullet and causing death by obstruction. Minski (1895) says, further, that in 1763 Dallas and Monroe snared a pedunculated one which a man of 64 ejected into his mouth; but that he died two years later of starvation owing to a recurrence. In 1936 when the literature was last reviewed in the British Journal of Surgery, J. D. Rose found only 49 cases, and all of them seen at autopsy, while up to 1943 Adams and Hoover (1945) were able to collect some 91 benign œsophageal tumours although only 6 had been clearly identified as leiomyomas. When Calmenson and Clagett, however, in 1946 examined 44 such innocent growths in 7459 post-mortem specimens at the Mayo Clinic 32 of them (or 73 per cent) proved to be leiomyomata: while Harrington and Moersch (1944) working at the same clinic a few years earlier had identified only 15 benign neoplasms in 11,000 patients suffering from dysphagia. Schafer and Kittle (1947) likewise found 11 during 6001 autopsies up to 1947. Chi and Adams (1950), reviewing the records of the clinics of the University of Chicago, discovered 2 innocent, compared with 246 malignant, tumours of the œsophagus occurring between 1936 and 1947. Dugan and Meagher, writing in 1950, added a further 14 instances to the total. Up till then, in short, the entire literature afforded less than 200 innocent œsophageal tumours, and all but about 25 of them were found at autopsy: of these the great majority were mucosal or submucosal in type. By contrast, and perhaps significantly, in the past two years another 12 leiomyomas of the œsophagus have been described in the medical press, and of these most were intramural and presented as mediastinal rather than as tumours of the œsophageal lumen. (Cornell, Shehadi and Sharnoff (1950) and Garlick and Stegmaier (1951).)

Innocent growths of the mucosal or submucosal type as they grow project into the lumen, and are therefore affected by peristaltic action, being drawn more and more away from the wall of the œsophagus until their attachment becomes attenuated into a pedicle, sometimes long enough to allow a spectacular regurgitation into the patient's mouth. Such tumours arise mostly just below the level of the cricoid, or in the lower third of the œsophagus, and comprise polypi, papillomas, adenomas, fibromas, epithelial cysts, lipomas, and some leiomyomas. Since they obstruct the lumen they are most likely to cause dysphagia as a presenting symptom; or bleed because they ulcerate.

Intramural benign tumours, on the other hand, rarely cause dysphagia because they do not usually cause a stricture of the lumen as does a carcinoma, even when they almost completely encircle it. The mucous membrane over them often ulcerates, however, giving rise to obscure but occasionally quite massive hæmorrhages into the bowel; or, as their bulk increases, they may press upon an adjacent bronchus and cause collapse of a lower lobe, or a pleural effusion. The great majority of these growths are leiomyomas, arising in the lower third of the œsophagus and extending in the plane between circular and longitudinal muscle coats. Sometimes they appear multicentric or diffuse in form, involving large segments, but characteristically they are circumscribed even when large, and diffuseness must arouse suspicion of leiomyosarcoma. Of 9 intramural growths described by Harrington (1949) 7 were leiomyomas and 2 leiomyosarcomas. He suggested that innocent growths did not cause ulceration of the mucosa, and that bleeding was therefore evidence of malignancy, but this is certainly incorrect.

Radiologically the leiomyoma may show as a lobulated opacity in the posterior mediastinum, overshadowed in anteroposterior views by the heart. Once distinguished, usually by an oblique picture, it may be seen to move on deglutition or peristalsis. During a barium swallow the mucosal folds over the tumour are ironed out, but those opposite to it are normal; and the flow of barium is generally unimpeded past the tumour since the other wall dilates. Madden and Olmstead (1950) have also pointed out that a helpful feature is the distinct shelf formed where the normal œsophageal wall meets the rigid segment involved by growth. This is best seen in profile view, and serves to differentiate intramural from extrinsic mediastinal tumours pressing from without, since in the latter the point of impingement is generally a smooth convexity and not a sharp angle.

Chest X rays may also show an associated pulmonary collapse or pleural effusion.

Œsophagoscopy for intramural leiomyomas is often negative, the instrument passing the growth effortlessly as does swallowed barium; and since

it is essentially mobile its presence is not felt. But if the tumour is distinguished, or the scars of previous ulceration seen on the mucosa covering it, it is important in view of the surgical measures indicated *not* to perform a biopsy and so impair the integrity of the mucosa.

The treatment of choice is enucleation of the leiomyoma. Since the growth is encapsulated this presents no technical difficulty providing that the correct diagnosis has been made; and the surgical risks involved are minimal. The principal error to be avoided is a quite unnecessary resection of the œsophagus, which carries with it even to-day a substantial mortality. Similarly every possible precaution must be taken that the lumen of the gullet is not breached. To facilitate this a stomach tube is passed which can readily be felt, marking the mucosal canal and providing a valuable guide to the dissection.

FIG. 288.—An overpenetrated view of the mediastinum in which a rounded opacity can be seen behind the right border of the heart and overshadowed by it.

CASE REPORT

A man aged 45 suffered from hæmatemesis and melæna two years prior to being admitted to the care of Dr. John Kidd at Amersham General Hospital with a further severe attack of hæmatemesis with melæna at the beginning of 1952. There had been no previous indigestion, but the first hæmatemesis was preceded by three days of pain in the chest; and there was no digestive disturbance of any kind between the attacks.

On admission he was exsanguinated and his hæmoglobin 26 per cent. Recovery was rapid except that after some weeks he developed pain in the left chest, accompanied by a pleural rub and followed by an effusion. A radiograph of the chest showed partial atelectasis of the left lower lobe and a small effusion. (*Fig.* 288.) Occult blood tests were soon negative, but became positive for a short time without signs of frank hæmorrhage.

He slowly recovered and had no indigestion. Barium meals were quite negative, showing no sign of any gastric or duodenal abnormality. Dr. Crichlow, the radiologist, remarked, however, upon an abnormal rounded opacity behind the heart shadow which he detected during screening of the chest, and which could be seen only from certain angles. Repeating the barium swallow he found slight dilatation of the œsophagus in its upper and middle thirds, and at the junction of middle and lower, where it abutted on the tumour, the barium formed a distinct angle or 'shelf' (*Fig.* 289). Otherwise it passed normally into the stomach without delay and without obvious interference with the mucosal pattern. By this time the patient seemed completely restored to health, and was referred to the London Hospital in September, 1952, for further investigation of his intrathoracic lesion.

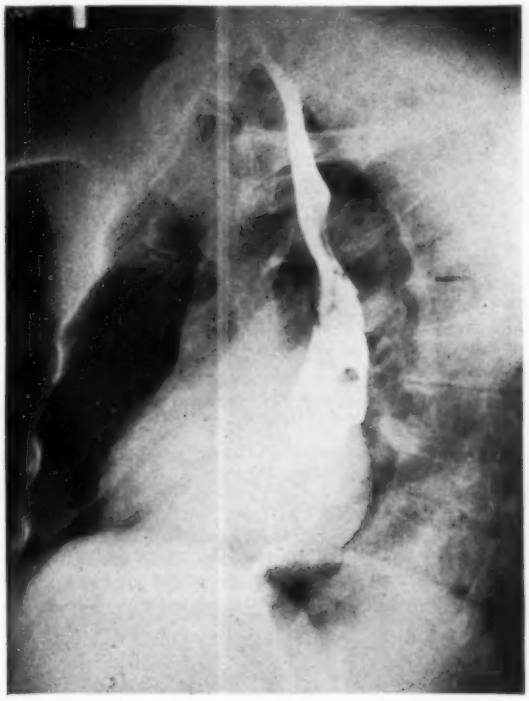

FIG. 289.—Barium swallow showing 'shelf' formation at the junction of middle and lower thirds of the œsophagus.

The barium investigations were now repeated by Dr. Murray and again no trace of peptic ulceration was found; but the shelf at the lower end of the œsophagus was distinct and the opposite wall of the lumen could be seen to dilate as the barium flowed over the rotundity of the tumour. At no time, however, was its demonstration easy.

Œsophagoscopy showed some dilatation of the upper two-thirds, but the instrument passed into the stomach at 40 cm. without hindrance, and no tumour could be felt or seen. The mucosa was normal except for a small scar on the posterior wall of the lower third.

It now seemed likely that an intrinsic innocent tumour of the œsophagus was present to which the previous episodes might be related; and of such tumours the commonest is the leiomyoma.

A right thoracotomy was proceeded with, the chest being entered through the bed of the resected seventh rib. A lobulated and roughly fusiform mass could at once be seen occupying most of the lower third of the œsophagus, the longitudinal muscle-fibres of which enveloped it. As the surface of the growth itself could not be seen, and its nature, though not its formidable

proportions, remained in doubt, the œsophagus was mobilized from the mediastinum and tapes passed around the normal portions proximal and distal to the tumour. The latter was now palpated and presented a curious almost completely encircled the œsophageal lumen. A few fine interrupted silk sutures to close the breach in the longitudinal coat completed the operation, and the chest was closed.

A B

FIG. 290.—A, Specimen after removal. The lumen of the œsophagus with the circular muscle coats passed through the gap in the middle and was almost completely encircled by the growth. B, Specimen extended. The scale is in centimetres.

nobbly mobility beneath its outer sheath of muscle, rather like feeling a bagful of knuckle-bones, and totally unlike a carcinoma. The longitudinal coat was then incised and at once a pallid, smooth, encapsulated surface presented. Around it a plane of cleavage was readily

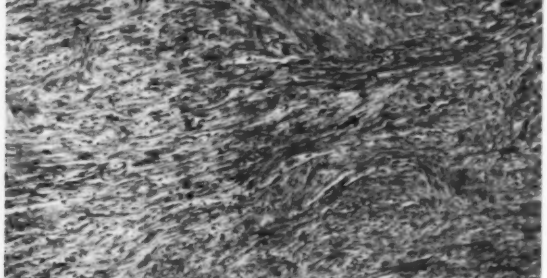

FIG. 291.—Leiomyoma. (× 42.)

established, partly by stroking away the œsophageal muscles with a dissector swab, partly by sharp dissection, and little by little the lobulated mass of a leiomyoma was delivered, arising principally, it appeared, between circular and longitudinal muscle coats. At this point a stomach tube was passed down the œsophagus by the anæsthetist, traversing the area of the tumour without hindrance, and thereafter providing a most valuable guide to dissection and enabling the now 'splinted' mucosal canal to be peeled safely away from the leiomyoma without risk of accidental puncture. As the whole growth was delivered it became clear that it had in fact

The patient made an uninterrupted recovery and there has been no recurrence either of lung symptoms or of intestinal bleeding.

The removed tumour lay naturally in circular form, with its extremities almost touching, and through this ring had passed the mucosal lumen and circular fibres of the œsophagus. It was lobulated, parchment-white in colour, and rubbery in consistency. When unfolded, its length was some 15 cm. and its breadth at the widest point 5·5 cm. (*Fig.* 290).

Histologically the appearances were of a typical leiomyoma, well-encapsulated, and with no trace of malignant change. The stroma was made up of a rich fasciculation of bundles of smooth muscle-fibres with occasional fibrous tissue septæ (*Fig.* 291).

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CARCINOMA OF THE FLOOR OF THE MOUTH

By A. J. GARDHAM

This article is based on information collected for a contribution to the discussion on carcinoma of the floor of the mouth at the meeting of the Association of Surgeons at Cardiff in April, 1952. The information is based on the records of 72 patients treated at University College Hospital. Many of them were treated by my colleagues, to whom I am very grateful for allowing me to make use of the case records. The series includes a patient treated by Christopher Heath in the year 1875 (*Lancet*, 1899, I, 1354) because this case shows the possibilities of operation in unfavourable circumstances. The rest of the patients were treated between 1920 and 1947 and the series includes all cases which it has been possible to trace. Microscopical proof of the nature of the lesion is lacking in some cases, particularly in those treated in the early days of interstitial irradiation, but I have studied the records very carefully and I think the series may be taken to represent fairly the results of treatment of carcinoma of the floor of the mouth by various methods at a London teaching hospital over a period of twenty-seven years.

RESULTS OF TREATMENT

The series is unsuitable for detailed statistical analysis but two important facts emerge from the study of the records. The first concerns the curability of the disease. Between 27 and 28 per cent of the patients treated at University College Hospital lived more than five years and are either still living free from recurrence or have died of other diseases after the five-year period. This gives an overall cure-rate which corresponds very closely indeed with those quoted by Sir Stanford Cade and Professor Windeyer. Professor Windeyer's cases were treated by two different techniques one of which he believes to have been superior to the other, but the cure-rate is practically the same. The patients in University College Hospital were treated by many different techniques: some by surgery, some by irradiation, and some by a combination of the two methods. The fact that the overall cure-rate is so constant with different forms of treatment suggests very strongly that there is a proportion of cases of carcinoma of the floor of the mouth in which the disease is relatively benign and localized and can be cured by any one of a number of different treatments.

The second significant fact which is shown by the case records from University College Hospital is the extreme rarity of good palliative results. In this series of 72 cases there are 20 cures and 8 patients who died of other causes within five years of their treatment. We have therefore been able to secure disappearance of the growth in 38 to 39 per cent of the patients treated; 54 per cent were complete failures in which there is no evidence that the patient was benefited by the treatment given. In the whole series of 72 there were only 5 patients who were not cured who survived more than 2 years. These figures suggest that palliation is so rarely achieved that it is not an object worth aiming at. This is a fundamental fact which is of great importance in deciding to what lengths one should go in an attempt to cure the disease.

The fact that the cure-rate is so constant with methods differing as widely as those used by Sir Stanford Cade, Professor Windeyer, and the various members of the staff at University College Hospital, suggests very strongly that there is little to be gained by comparing the percentage of cures obtainable by the various methods of treatment. The convention of breaking down the total mass of cases into stages is also of very doubtful value. The impossibility of preselecting favourable cases in any form of malignant disease is clearly demonstrated by the cure-rate claimed in so-called 'early' or 'stage I' cases. If it were possible to distinguish clinically or microscopically between a localized growth and a spreading growth and between a growth of high malignancy and a growth of low malignancy there would be no difficulty in producing a series of cases of malignant disease in which the cure-rate is 100 per cent. The fact that no such series exists demonstrates that neither clinical nor microscopical examination of a growth gives a reliable indication of its malignancy and the imperfection of judgements based on any means of assessment now available can be measured by the amount by which the cure-rate in so-called 'early' cases falls short of 100 per cent. In carcinoma of the floor of the mouth 'early' is a particularly misleading term. The fact that a growth has existed for a short time gives no information regarding its malignancy and very little concerning its prognosis. It is proposed in this article to group the cases simply as successes and failures and to consider the application of treatment to each of these groups. The cases which are capable of palliation are too few in number to make a separate group.

Successful Cases.—If the 8 patients who died of other causes within five years of their treatment are excluded, 20 patients from the University College Hospital series fall into the curable group. The impossibility of preselecting this group has already been stressed, and in the series on which this article is based the 'curable' cases showed no common factor in the history or physical signs. It is possible that preselection by histological methods will assume a greater importance in the future.* Histological investigation in the series at present under consideration was not sufficiently detailed to be of any value from this point of view and no other common factor can be found among the cases which go to make up the curable group.

Of the cured, 10 were treated by operation—5 by interstitial irradiation alone, 2 by interstitial irradiation followed by gland operation, and 3 with tele-irradiation. Neither of the 2 cases treated by

* Wood, Constance C. E., and Boag, J. W., "Researches on Radiotherapy of Oral Cancer", *Medical Research Council Report No.* 267, p. 67.

interstitial irradiation and gland dissection proved to have microscopical evidence of growth in the glands and the credit for these 2 cases must go to irradiation. Thus, exactly 50 per cent of these cases were cured by operation and 50 per cent by some form of irradiation. For a comparison of the merits of these two methods, the years between 1925 and 1933 may be taken because operation and irradiation were used at University College Hospital to about an equal extent during this period. Most of the operations were done by Wilfrid Trotter. Most of the irradiation was done by me by the interstitial method. During this period 12 patients were cured; 6 by operation and 6 by irradiation, though 2 of the irradiated cases subsequently had a gland dissection performed. The equality of the numbers serves to strengthen the suggestion that it is the nature of the disease and not the nature of the treatment which decides the possibility of cure, but there is a further significant finding which led me to abandon interstitial irradiation in the treatment of growths which I believe to be curable: 2 of the 5 patients cured by interstitial irradiation died of growths in other parts of the mouth. One developed a sarcoma in the irradiated area but was apparently cured by surgical removal of this lesion. One patient developed an extensive sequestration of the lower jaw seven years after treatment and only survived as a result of his own determination through a period of two years' agonizing pain. A little further on in the records there is a third case cured of carcinoma of the floor of the mouth by interstitial irradiation who died of an independent primary growth in the mouth eight years later. Thus, of 10 patients in the whole series who were cured by irradiation, 3 died of second primary growths, 1 required operation for a sarcoma in the irradiated area, and 1 underwent a great deal of misery as a result of a necrosis of the lower jaw. These 5 were all personal cases treated by interstitial irradiation and there is no evidence in our figures to show that tele-irradiation carries the same dangers though there are published figures* relating to growths in other situations which suggest that this is the case.

All the patients who were treated by operation had some deformity as the result of the operation but not one of them was sufficiently bothered by the deformity or by the disability which followed his primary operation to require a plastic repair, though the average duration of life after operation was fifteen years. There is no record of any case cured by operation in which an independent primary growth developed subsequently. On the above evidence there are some grounds for preferring operation to irradiation in cases in which the prospects of cure are good, but the evidence does not justify a final conclusion. The question of operative mortality is of course important in this connexion. The records of cases operated on since the introduction of chemotherapy suggest that this is no longer a factor of much importance, but the number of cases is still too small to justify conclusions. On the other hand it is a pleasing thought that the disadvantages of irradiation in the cases quoted are concerned with side-effects and not with the power of irradiation to cure the primary lesion. Some observers hold the view that further technical improvements in the application of irradiation are unlikely to increase the percentage of cures, but it may well be that the technical improvements already in use, or those still to come, will reduce the side-effects to a point which makes it clear that for the curable lesion irradiation is better than operation. The conclusion on this point must await the results of a further ten or fifteen years' experience.

Failures.—If it can be accepted that apart from the side-effects there is little to choose between irradiation and operation in favourable cases it is more profitable to consider the failures in the present series with a view to deciding whether it is possible to separate from this group some which should have been irradiated and others which should have been submitted to operation.

Of the cases in the University College Hospital series 54 per cent were failures. All but 2 of them received treatment of some sort. The average survival of the 2 untreated cases is thirteen months compared with ten months in the treated cases and there is no evidence that the patients in this group who were treated were helped in any way. There is evidence that some of them were made worse, because the records contain frequent references to pain of a very severe degree and occasional references to extensive bone necrosis. There were 39 patients in this group. One died as a result of dental extractions, 2 were treated by surgery in the first instance and 2 others were operated on after radiotherapy had failed; 34 of them were treated primarily by irradiation. Most of these patients were referred to the radiotherapy department by surgeons. This may be taken to indicate that the surgeons who referred the cases rated the possibility of cure by operation very low. It may be that they were right in doing so, but there is no evidence that the cases rejected by surgeons as unsuitable for operation stood any chance of cure or reasonable palliation by irradiation though they could not well be turned away from the department and there is therefore a very strong case for reconsidering the possibilities of surgery in this group.

I have tried to classify the 39 patients who made up our group of failures as 'operable' or 'inoperable'. I have classified as 'operable' those in whom it would have been possible to remove all visible and palpable extensions of the growth by an operation of the type which can now be used without any significant operative mortality, and as 'inoperable' those with distant metastases, those with local extensions which were irremovable or only removable at the cost of producing an intolerable deformity, and those who for reasons which I give below I should myself reject for operation. On this classification 16 of the 39 were 'operable', 8 were doubtfully 'operable', and 15 were 'inoperable'. Whether any of the 'operable' cases would, in fact, have been cured by operation will never be known, but it is certain that none were cured by other means of treatment.

Selection of cases to be submitted to operation in this group is important, but the selection should not be too rigid. The recurrence rate as far as can

* Baclesse, F. (1949), *J. Amer. med. Ass.*, **140**, 525.

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be ascertained in the cases operated on at University College Hospital was only 27 per cent. This figure would have been very difficult to attain if operation had been offered to every patient in the group of failures whom it might have helped, and suggests that operation has not been used sufficiently often.

It must be accepted that some cases are beyond the help of surgery when they are first seen. There are certain well-defined contra-indications to operation. The most absolute of these is the presence of extensive involvement of glands when the primary growth is still small. In such cases operation is bound to involve division of lymphatics which contain active growth and is very likely to be followed by widespread recurrence in the wound. Growths in which the submucous spread is very wide in comparison with the size of the ulcer also do badly with operation, perhaps because the submucous spread is a sign of widespread lymphatic invasion. A third group in which surgery offers little hope is that in which the bone of the lower jaw is expanded. The clinical picture in these cases is different from that which is presented when the bone is involved from outside, and it is probable that the expansion of the bone results from invasion by lymphatic spread along the vessels to the interior of the bone and is again a sign of lymphatic invasion disproportionate to the extent of the primary growth. Growths which are soft and do not show any signs of fixation to surrounding tissues may also be very highly malignant and therefore unsuitable for surgery. The lack of fixity in some cases is apparently due to the fact that the epithelial portion of the new growth is very active and is not accompanied by fibrosis. Experience shows that growths which are very mobile and therefore apparently present the surgeon with an easy technical problem often give bad results if they are submitted to operation.

On the other hand local extensions of the growth beyond its point of origin seldom make it unsuitable for operation. Involvement of the anterior part of the tongue is nearly always present and is certainly not a contra-indication to operation. Direct spread to the cheek or lip adds very little to the difficulty or severity of the operation and is certainly not a contra-indication. Attachment to the surface of the jaw is nearly always present. It has none of the evil significance which is associated with expansion of the jaw and neither the immediate nor the remote results of surgery are affected adversely by removal of half or more of the lower jaw in continuity with the growth. On the whole operation is seldom contra-indicated by the size of the growth provided that the rate of progress has not been unduly fast and often a big growth showing widespread attachment is more favourable for surgical treatment than a smaller and more mobile growth which has progressed with greater rapidity.

Christopher Heath's Case.—This case is quoted in some detail because it is an excellent example of the possibilities of surgery in unfavourable circumstances. Particulars of the case are as follows :—

The growth had existed for twenty months before the patient was treated at University College Hospital. An inadequate excision had been done elsewhere five months before Heath operated. The operation was carried out without the help of modern anæsthesia, and of course without the help of antibiotics, in September, 1875. Heath himself describes the operation as follows : "The tongue, centre of the jaw, and all the sublingual tissues were removed by dividing the skin in the middle line then sawing the jaw through on either side and enclosing the whole of the disease with the wire of the galvanic ecraseur". The diagnosis was confirmed by microscopical examination and the specimen is still in

FIG. 292.—Specimen from the Museum of University College Hospital showing the tissues removed by Christopher Heath in 1875. (24-year survival.) (× ½.)

the museum of University College Hospital. It is illustrated in *Fig.* 292, which shows that it consists of part of the lower jaw including the portion between the mental foraminæ and extending about ½ in. behind one of them. Behind this lies an ulcerated surface which extends from the teeth over the whole of the floor of the mouth in the incisor region and on to the under-surface of the tongue on the right side. It was this specimen which first drew my attention to the possibilities of surgery in apparently unfavourable cases, for this patient lived 24 years and died of other causes in 1899.

Heath's operation owed some, at least, of its success to the observation of two fundamental rules in the treatment of malignant disease. The first is that the diseased tissue should be removed without regard to subsequent repair. The second principle involves recognition of the fact that growths in the mouth do not spread only along mucous surfaces, and that consequently the best operation for malignant disease is one which removes a sphere of tissue with the point of origin of the growth at its centre. This is the basis for removal of the primary lesion and its immediate extensions. It may, of course, require to be extended in any direction in order to clear the lymphatic fields.

Figs. 293–295 show the application of the second principle to carcinoma of the floor of the mouth and are sufficient to explain why it is that operations performed from within the mouth on the assumption that the growth spreads in relation to mucous surfaces only are so seldom adequate.

Heath's operation really had most of the essentials in that it removed the anterior part of the tongue and an adequate portion of the alveolus, and the principle of the present-day operation is the same.

THE PRESENT-DAY OPERATION

The patient is admitted to hospital a few days before operation. Remaining teeth are cleansed but no extractions are made. Treatment with penicillin and streptomycin or terramycin is commenced twenty-four hours before operation. The mouth is cleansed with simple mouth-washes and the patient is taught,

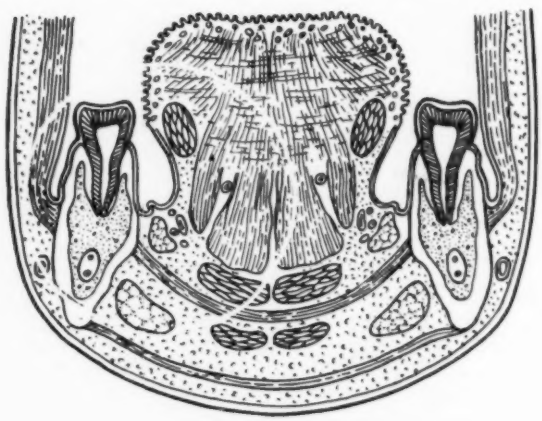

FIG. 293.

of the horizontal ramus of the lower jaw, and unless the growth is very small indeed the portion removed should include the whole thickness of the jaw in order to reduce the size of the floor of the mouth and to make closure easier. In extensive growths it is also necessary to remove the whole or part of the ascending ramus.

The operation is performed as follows: An incision is made from the middle of one mastoid process downwards along the anterior border of the sternomastoid to cross the neck at the level of the hyoid and run upwards again to the opposite mastoid process. The flap marked out by this incision is turned up. In most cases the external jugular veins are divided in the line of the incision and turned up with the platysma in the flap. If the glands of the neck are obviously involved it is probably better to include skin only in the flap. In a few cases the skin is actually involved on the outer side of the jaw where the growth has spread out into the alveolobuccal sulcus and thence into the cheek. In these circumstances the incision must be modified, but it should always be planned to expose both submaxillary triangles adequately and to allow a clear view of the side of the tongue from the hyoid to the tip when the jaw has been divided. The lower poles of the

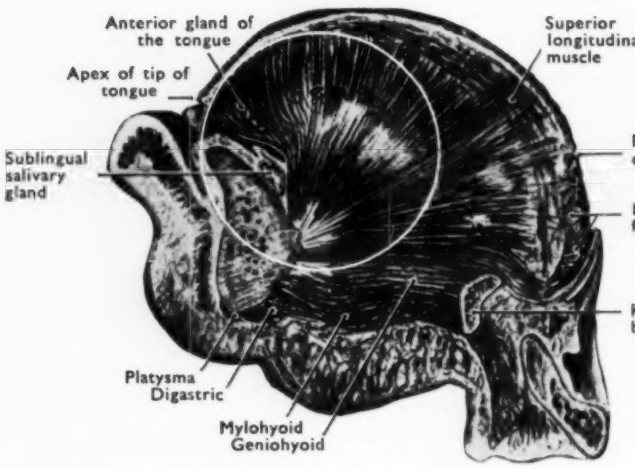

FIG. 294.

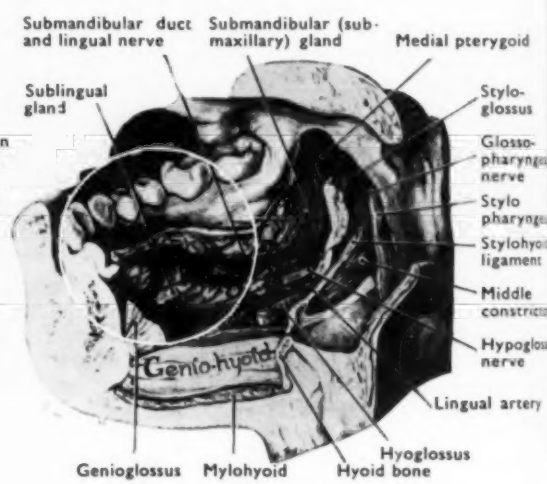

FIG. 295.

FIGS. 293–295.—Diagrams to show the anatomical basis of operations for carcinoma of the mouth. The point of origin of the growth is at the centre of the circle in each of the illustrations.

as far as possible, to use a feeding cup with a rubber tube attached to its spout.

The patient is anæsthetized and a cuffed intratracheal tube is passed and the rubber bag inflated to stop aspiration round the tube. In addition to this the pharynx is lightly plugged with gauze.

The next step is to examine the growth fully as it is seldom possible to appreciate its full extent until the patient is under an anæsthetic. This is a most important step as the success of the operation depends entirely on a full and accurate knowledge of the exact situation and extent of the growth.

The commencing steps of the operation are more or less standardized as the lesion is always close to the midline, and it must be assumed that the lymphatics of both submaxillary triangles are involved. It is advisable in all cases to remove some portion

parotid glands are turned upwards with the skin-flap and in front of this the flap is turned up to a point about $\frac{1}{2}$ in. above the inferior margin of the jaw. The facial vessels are isolated and divided $\frac{3}{4}$ in. above the point where they cross the lower jaw as the gland which lies on the outer surface of the jaw at this point may be involved.

A routine dissection of both submaxillary triangles is then made from behind forwards. This is commenced by pulling the sternomastoid backwards and exposing the digastric muscle and ligaturing the facial vein close to the internal jugular. The lymphatic gland which lies at the junction of the veins is often enlarged and it is wise to remove it and send it separately for microscopical examination as this gives a valuable indication of the extent and direction of the lymphatic spread. The external carotid

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artery is generally ligatured in continuity close to its origin on both sides. This step was originally taken by Trotter as a precaution against secondary hæmorrhage; for this purpose it is now unnecessary, but the diminished bleeding makes it easier to complete the operation with accuracy. I do not remove the stylohyoid or the posterior belly of the digastric as I think they play an important part in the mechanism of swallowing. The submaxillary salivary glands are then freed posteriorly by dividing the facial artery close to its origin and dividing and ligating the band of tissue which connects the posterior aspect of the submaxillary gland with the parotid. More anteriorly the mylohyoid is divided just above and parallel to the hyoid and turned upwards to complete the exposure of the deep portion of the submaxillary salivary gland.

When this part of the operation has been performed on both sides the field is ready for division of the jaw and exposure of the primary growth. The technical steps of the operation from this point differ very little according to the amount of the horizontal ramus to be removed but slight variations are necessary if the growth extends on to or close to the ascending ramus. When this is the case and it is necessary to remove the portion of the ascending ramus which lies in close relation to the mucous membrane I use the following procedure: With the diathermy cutting current the masseter is incised along the posterior border of the ascending ramus of the jaw up to a point just in front of the ear. Still using diathermy the muscle is freed from the underlying bone, working forwards and upwards until it is possible to insert the curved end of a cleft-palate elevator into the mandibular notch. Using this elevator to retract the soft tissues the ascending ramus is divided vertically rather less than a finger's breadth in front of its posterior border (*Fig.* 301 A). This division is best effected with a mechanical saw. A narrow strip of ascending ramus from the condyle to the angle is thus formed. This strip of jaw performs a very useful function subsequently as it angulates sharply forward and when the wound is healed occupies a position which is almost horizontal. Lying in this position it replaces the greater part of the horizontal ramus and even when the anterior division has been made to the opposite side of the midline the parts generally fall together sufficiently to give reasonably good function, though the union between them is fibrous and not bony. Once this strip of the ascending ramus has been freed from the rest of the jaw it can be drawn backwards with a bone hook to expose the pterygoid region. Bleeding is generally rather free from the pterygoid veins, and from the branches of the internal maxillary artery, in particular the

A

B

C

FIG. 296.—A, Tissues removed at operation for an extensive growth on the left side. The ascending ramus of the jaw appears to the left of the photograph and the tip of the tongue to the right. Beneath this the jaw has been divided at the symphysis. The centre of the photograph shows the muscle in the middle of the tongue and behind this lie the contents of the submaxillary triangle. B and C, Appearance of patient 8 days after operation. The patient's age is 77. A temporary tracheotomy and gastrostomy were done at the same time as the main operation. (Recent case.)

inferior alveolar artery which is necessarily divided, but the bleeding is not difficult to stop.

If the growth is sufficiently remote from the ascending ramus to make it possible to divide the jaw more anteriorly the foregoing step is unnecessary and the jaw is divided with a mechanical saw about ½ in. behind the attachments of the growth. The jaw is then divided anteriorly ½ in. in front of the growth or, if necessary, on the opposite side of the symphysis. It is very often convenient at this point to remove the isolated portion of the jaw. This is theoretically undesirable but it makes accuracy in

FIG. 297.—Sketch, made at time of operation, of portion of jaw removed in continuity with growth and contents of submaxillary triangle.

the ensuing part of the operation easy and probably has few disadvantages.

The next step is to incise the mucous membrane so as to display in the wound the surface of the growth. Generally the incision can be made in the lower alveolobuccal sulcus in the whole length of the bed from which the isolated piece of jaw has been removed, but if the growth has extended up on to the mucosa of the cheek the incision must be made at a higher level. The incision crosses the alveolus about ¾ in. behind the posterior margin of the growth and anteriorly crosses the midline to expose the frænum of the tongue and the floor of the mouth on the less affected side.

The growth is now displayed in its full relation to the tongue and it will often be found that it infiltrates the tongue to a much greater extent than was appreciable before the jaw was removed. It remains only to remove a sphere of tissue with the point of origin of the growth at its centre in continuity with the contents of one or both submaxillary triangles and the periosteum of the sector of jaw which has been removed. If the growth is very strictly unilateral it may be justifiable to remove the contents of the opposite submaxillary triangle separately, but generally the sphere includes the orifice of the submaxillary duct on the less affected side and the tissue removed must therefore be in continuity with the contents of both submaxillary triangles. This portion of the operation must be performed deliberately and is best done with cutting diathermy, picking up the margins of the tissue which is to be allowed to remain with thread tractor stitches as the dissection proceeds. Branches of the lingual artery generally bleed freely in spite of the ligature of the external carotid. The main trunk of the lingual nerve on the more affected side is necessarily divided. In most cases the main trunk of the hypoglossal is spared on both sides but no attempt is made to spare any of its branches in the substance of the tongue. Most growths extend upwards on the surface of the hyoglossus on the more affected side and it is wise to carry the incision at least as far as the midline of the tongue immediately above the site of the lesion. If the lesion is a reasonably

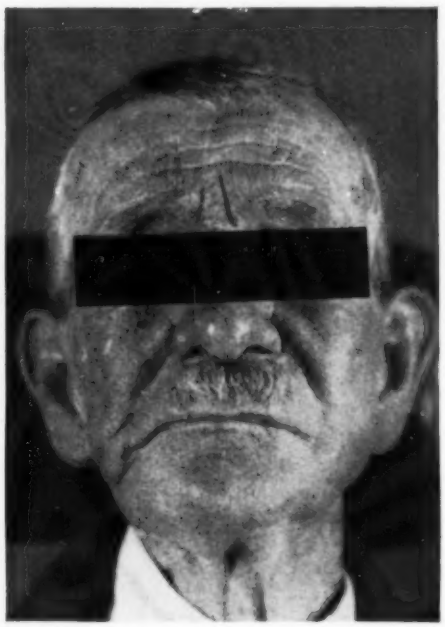


FIG. 298.—Appearance of patient (same case as *Fig.* 297) 7 years later. There has been no plastic repair. The deformity in this case is less than the average.

small one it is possible to isolate the sphere of tissue behind and in front of the site of the lesion by cutting into the muscle substance of the tongue in a plane more or less parallel to its mucous surface thus coring out a block of tissue and leaving a very valuable area of well-nourished mucous membrane to form the new floor of the mouth. *Fig.* 299 B shows the block of tissue removed in a typical case.

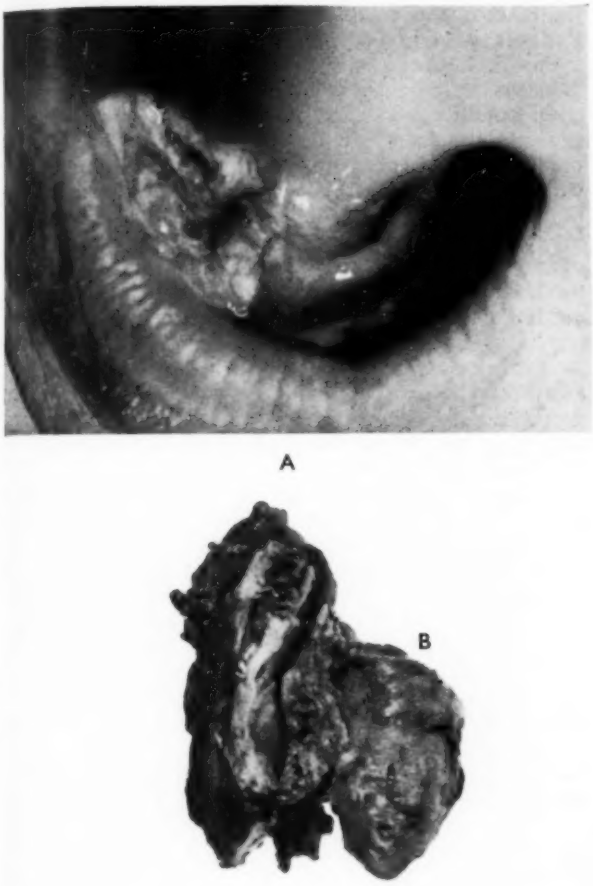
The excision is now complete and repair may be considered. Generally the only points of difficulty are immediately adjacent to the saw-cut through the bone of the horizontal ramus. Closure is easier when a considerable portion of the jaw has been removed and is very simple when a portion of the ascending ramus has been removed. The first step is to free the mucosa very gently from the bone for about ½ in. away from the saw-cut. A decision is then made as to whether any attempt shall be made to unite the remaining portions of the jaw. It matters very little whether or not the jaw is wired and there is certainly no point in wiring unless the fragments fall together easily. Having wired the jaw if it appears desirable to do so it is generally best to cover the main wound with gauze pads and to start working from within the mouth. It is very easy to obtain an adequate view through the mouth at this stage and one is less liable to leave an unnecessary deformity. I do not aim at very close apposition of the edges of the mucous membrane but prefer to bring broader raw surfaces together with interrupted catgut sutures. The details of the closure vary, but

there is seldom any difficulty in providing a covering for the floor of the mouth. It is well not to deform the base of the tongue unduly as this makes for greater difficulty in swallowing subsequently and it is desirable to leave some portion of the tip of the tongue mobile. This is not essential for speech but a patient with no mobile tip to his tongue may undergo a great deal of embarrassment subsequently as a result of dribbling and a very small mobile segment is sufficient to prevent this. The portion of the tongue between the base and the tip may be freely used to reconstruct the floor of the mouth and can usually be made to fill the gap without the least difficulty.

When the closure has been satisfactorily completed from within the mouth, gloves and instruments are changed and the external portion of the operation is concluded. The first step is to reinforce the reconstructed floor of the mouth by drawing muscle tissue together where possible beneath the sutured mucosa. If there is a point where closure is imperfect and muscular reinforcement is impossible it is better to support it with a 'Vaseline' plug and to leave a corresponding portion of the skin wound unsutured. This practice often leads to a fistula, but such fistulæ are often small and are rarely accompanied by any considerable degree of sepsis in the neck.

The skin wound is now sutured with interrupted thread stitches, leaving a sufficient gap for exit of the 'Vaseline' plug if one has been used. Tube drainage is of very doubtful value and most cases which do not require a 'Vaseline' plug are suitable for complete closure. (*Fig.* 296.)

It will be noted that this operation is designed to leave the lymph fields beneath the sternomastoids untouched and the posterior belly of the digastric muscle can be used in the manner which Trotter originally suggested to shut off the carotid sheath from the rest of the incision. It is occasionally necessary to clear the deep cervical lymphatic fields at a later stage, and some guide as to the necessity of this further operation can be obtained by microscopical examination of the jugulodigastric gland which is removed in the early stages of the main operation. Clearance of the deep cervical lymphatic fields after an operation for clearance of the submaxillary triangles is considerably more difficult than a routine gland dissection because of the fibrosis around the origin of the external carotid, but it is otherwise quite satisfactory.



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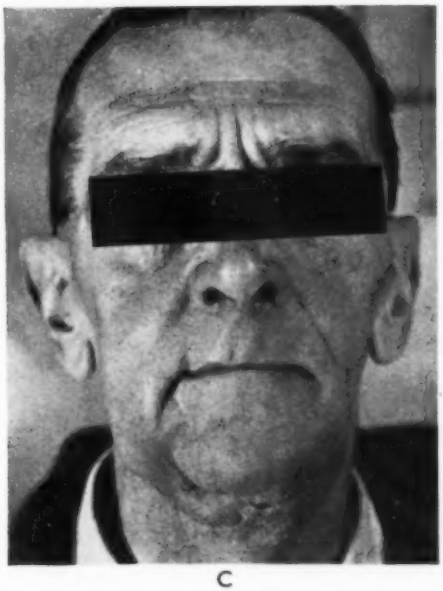


C

FIG. 299.—To show a typical case. A, Lesion before operation. The superficial extent of the growth is shown through the open mouth. The tongue is elevated and displaced to the left. B, The growth appears in the middle of the specimen. It is bounded on the other side by the jaw from the angle to a point just beyond the midline and on the inner side by the right half of the tongue. C, Appearance of patient 18 months after operation. There has been no plastic repair. The deformity is average. This patient died soon after of another disease. No recurrence was present.

Tracheotomy.—Modern anæsthesia has made the technique of operations on the mouth very much more simple than it was in the past. In the past most of the difficulties arose from venous engorgement and respiratory obstruction, but neither of these complications occurs to any extent with an anæsthetic efficiently given through a nasal intratracheal tube. This method of anæsthesia is preferable to the older one of giving the anæsthetic through a tracheotomy made under local anæsthesia before the main operation has begun because patients who have been anæsthetized for a long period through a fresh tracheotomy opening are more liable to develop post-operative chest complications than those who

have been anæsthetized through a nasal intratracheal tube. Respiratory obstruction is still a formidable complication, but it does not now happen at the time of the operation. The danger period to-day is after the intratracheal tube has been withdrawn and the patient has returned to the ward. Respiratory obstruction at this stage arises from a combination

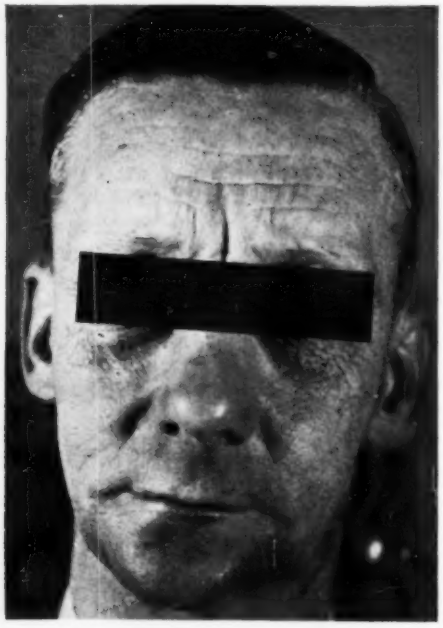


FIG. 300.—To show appearance 18 months after removal of contents of both submaxillary triangles and about 2½ in. of jaw extending to both sides of the midline. The soft-tissue tumour was large and the deformity is rather above the average. Patient remains well at 34 months.

of circumstances and it is by no means uncommon. A few minutes of grave respiratory obstruction at this stage may cost the patient his life because the effects of severe respiratory obstruction in elderly people are irreversible and lead to death in the ensuing forty-eight hours. For this reason I always do a temporary tracheotomy upon patients on whom I have operated for carcinoma of the floor of the mouth at the conclusion of the main operation even though at that time they are invariably breathing easily and without obstruction. I adopted this practice only a few years ago but I am quite sure of its value. The tracheotomy tube is withdrawn five to seven days after the operation and the fistula allowed to heal.

Gastrostomy.—Gastrostomy still has a real value in post-operative treatment of patients who have had major operations involving the mouth and pharynx. The operation is one which can be performed in a few minutes at the conclusion of the main operation and apart from the extra effort which it involves on the part of the surgeon it has very few disadvantages. The incision must be small in order that it may not discourage the patient from coughing in the post-operative period and the old-fashioned ink-bottle method of inserting the catheter is as satisfactory as any. I am inclined to think that gastrostomy should be used more widely than it is in these cases. The difficulties of swallowing in the first week or ten days after any major operation which involves the tongue and the floor of the mouth are very real. They are produced partly by œdema, partly by disturbances of the normal sensation of the mucous membranes, and partly by the mechanical interference with the normal mechanism for directing fluids into the œsophagus rather than the larynx. Patients vary very greatly in their ability to overcome these difficulties and a proportion of them begin to swallow with more or less ease within the first few days but the process of teaching them to swallow is a very laborious one and requires great skill and persistence on the part of the sister-in-charge and very considerable moral courage on the part of the patient. Repeated unsuccessful attempts at swallowing accompanied by inhalation of food and the distressing paroxysms of coughing which follow this are extremely exhausting and discouraging to the patient and I think it must be admitted that swallowing is re-established more easily and with less risk in patients who have been nourished through a gastrostomy opening in the early post-operative period and are only encouraged to practise swallowing when the initial disturbance of the operation is beginning to subside.

Septic Complications and Fistula Formation.—Local wound sepsis very rarely gives rise to any anxiety under modern conditions. Those who practised operations of this type before the days of chemotherapy and antibiotics will have no difficulty in remembering the horrors which spreading cellulitis of the tissue planes of the neck brought with it. I have not had a single case which has given me any anxiety from this cause since the end of the last war. The credit lies entirely with modern methods of combating sepsis and I cannot claim that improvements in operative technique have any connexion with it.

Fistula formation still occurs. It is generally possible to pick out those cases in which fistula formation is likely and to make provision for the formation of a harmless fistula by leaving the skin unsutured at a suitable point and inserting a 'Vaseline' plug between the weak point in the mucous membrane and the unsutured area in the skin. Such fistulæ are harmless. Many of them close spontaneously and those which do not can be closed without difficulty at a subsequent operation. It is always better to allow a fistula to form than to leave an insecurely sutured area in the mucous membrane leading into a closed cavity in the tissues of the neck.

Septic pulmonary complications still occur. They are more troublesome during convalescence than they are in the immediate post-operative period and they may prove serious or fatal as late as the third or fourth week. They occur most commonly in patients who have found it difficult to learn to swallow and are undernourished and discouraged. I think that the prolonged presence of an indwelling pharyngeal tube is a contributory factor and that gastrostomy is the best insurance against this form of septic complication.

Restoration of Function.—Most patients who suffer from carcinoma of the floor of the mouth are elderly, but their ultimate functional recovery even after the most extensive operations is surprisingly

good. I am satisfied if my patients can talk intelligibly, swallow solid food, and mix with others without being conscious of a gross deformity. Recovery of speech is one of the most pleasing aspects of operations for carcinoma of the floor of the mouth.

FIG. 301.—A, B, and C. To show maximal deformity. A, The tissues removed are shown from the outer side. The coronoid process can be seen in this specimen. The condyle has been left in situ with a strip of the posterior margin of the jaw. B, The tissues removed are seen from in front and below. On the left of the photograph the growth extends on to the cheek. From the cheek it passes across the lower jaw to infiltrate the hard palate, the tonsillar fossa, and the substance of the tongue. C and D, Appearance 2 years later. The deformity is maximal, but the patient, who is now aged 81, does not wish for any plastic repair. He eats and talks without difficulty and smokes heavily. Patient remains well at 3 years.

The observation that the anterior part of the tongue is not necessary for intelligible speech is not a new one, for Gibbon, in his *History of the Roman Empire*, comments on the rapidity and certainty with which the power of speech returned to the early Christians when attempts had been made to silence them by removing their tongues. Recovery is equally rapid when the operation is performed for better motives and, on an average, speech is very difficult for a week, indistinct for three months, and good enough for all ordinary purposes soon after this. I have not generally asked for the assistance of a speech therapist because the patients concerned are elderly and not very receptive of new ideas, but in suitable cases I am sure that the assistance of a speech therapist would be a very valuable aid. Up to the present I have never encountered a patient who failed to learn to talk intelligibly by his own efforts.

The recovery of function as regards feeding and swallowing is much more variable, and is more closely related to the extent of the operation. Bony union between the fragments of the lower jaw seldom occurs but a firm fibrous union gives the patient a

very powerful and efficient bite and many of my patients are happy to demonstrate their abilities in this direction on the examining finger of anyone who can be persuaded to take an interest in the matter. Lower dentures are a great difficulty, as the operation not only deforms the alveolus but also abolishes the sulcus between the alveolus and the remains of the tongue. A few of my patients have been fitted with lower dentures which they find satisfactory but most of them are happier without. Those in whom a portion of the ascending ramus has been removed are never able to wear lower dentures because the remaining strip of the ascending ramus angulates too sharply forwards. The worst results as regards swallowing occur in patients in whom it has been necessary to remove large portions of the tongue immediately above the hyoid. Fortunately this is seldom necessary in growths which originate in the floor of the mouth.

External deformity is surprisingly little and I do not think that I have ever heard a patient complain of it. There are a few precautions which must be taken at operation with a view to avoiding unnecessary deformity. The most important is to avoid division of either lip and to turn the soft tissues of the chin and lips off the jaw in an upward direction so that they provide an intact covering for the reconstructed jaw at the end of the operation. A small nodule of mobile tissue in the anterior part of the tongue should be preserved if possible to prevent dribbling. The amount of bone removed from the lower jaw has surprisingly little effect on the ultimate deformity which is seldom severe and never intolerable (*Figs.* 297–301).

No account of operative treatment of carcinoma of the floor of the mouth would be complete without reference to the efforts which these cases demand from the nursing staff. The credit for each successful case belongs at least as much to the nursing staff as to the surgeon and anæsthetist.

CONCLUSIONS

1. Carcinoma of the floor of the mouth is a clinical entity. The overall cure-rate recorded after various forms of treatment is very constant and it is suggested that a cure-rate of about 28 per cent can be obtained by any one of various forms of treatment. Cases of second primary growths occurring after interstitial irradiation are quoted.

2. The rarity of good palliative results is stressed and it is suggested that cases which appear to be unfavourable as a result of local extension are more suitable for operation than for irradiation. Clinical types unsuitable for operation are defined.

3. The technical steps of the operation are described.

4. The indications for tracheotomy and gastrostomy are discussed.

5. The post-operative and ultimate functional results are described.

CARCINOMA OF THE FLOOR OF THE MOUTH

By J. L. DOBBIE, Manchester

I must express my appreciation of the honour of your invitation to address you on behalf of the Christie Hospital and Holt Radium Institute of Manchester.

For many years we have regarded the floor of the mouth as accessible to the effective use of radium. I propose to describe briefly the methods, and to illustrate the results in a consecutive group of patients.

It is necessary to understand that in treating cancer of the mouth we separate the radium treatment of the primary from the treatment of secondary lymphatic extension, which, whenever possible, is entrusted to block dissection.

Each primary treatment represents the individual application of an appropriate technique to the patient treated, and the results I am going to show all relate to moulds and implants in nearly equal proportions—X rays we practically never apply in the front of the mouth.

METHODS

The double mould is one of the most characteristic methods of the Manchester centre—if it is not as frequently used elsewhere that must be, it seems to me, for reasons unconnected with its merits. Like other brachyradium methods it has the advantage of confining the effect of the radium to the minimum necessary volume, and, like other moulds, it is extremely controllable as regards area covered and dose delivered. It has the particular merit that invasion on to, and even into, the jaw can easily be included in an effective field of radiation. By this method, invasion of bone is no contra-indication to radium treatment, or necessary bar to cure, in spite of the contrary opinion sometimes expressed.

The general arrangement of a double mould for the floor of the mouth is illustrated in *Fig.* 302.

Disease with lateral and forward extension lends itself to this form of treatment, and sometimes extension backward into the tongue can also be included if the outer mould is replaced by a submental plane of implanted radium, as illustrated in *Fig.* 303.

In about half the cases, however, a satisfactory mould is not possible, and then implanted radium must be used. *Fig.* 304 shows an example of a volume type of implant. The needles are 4·5 cm. long and pass through the tongue into the floor of the mouth.

A third common method is the radon implant in gold seeds—particularly suitable for small and superficial lesions, and for the aged.

If these techniques can give results they have enormous advantages. They achieve their end by the excellent method of simply restoring the mouth to normal, without loss of tissue or need for reconstruction. Moulds need no surgical procedures,

and even a full needle implant is a simple proceeding compared with the surgical equivalent. Gold seed implants can give effective treatment to some patients

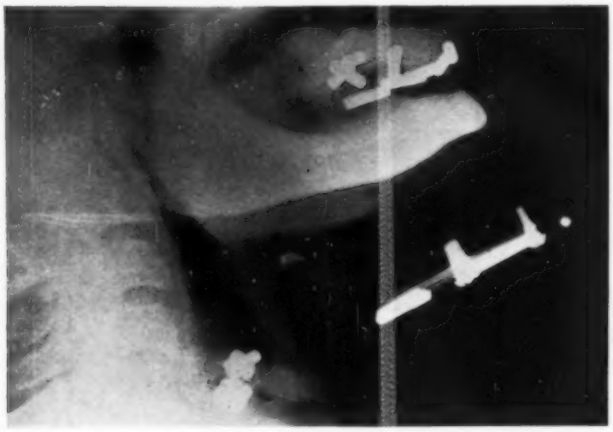


FIG. 302.—Double radium mould for floor of mouth.

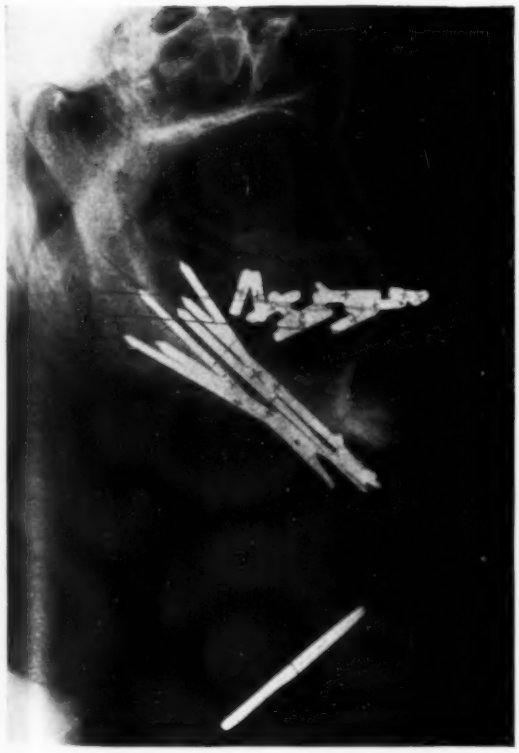


FIG. 303.—Outer mould replaced by implanted needles.

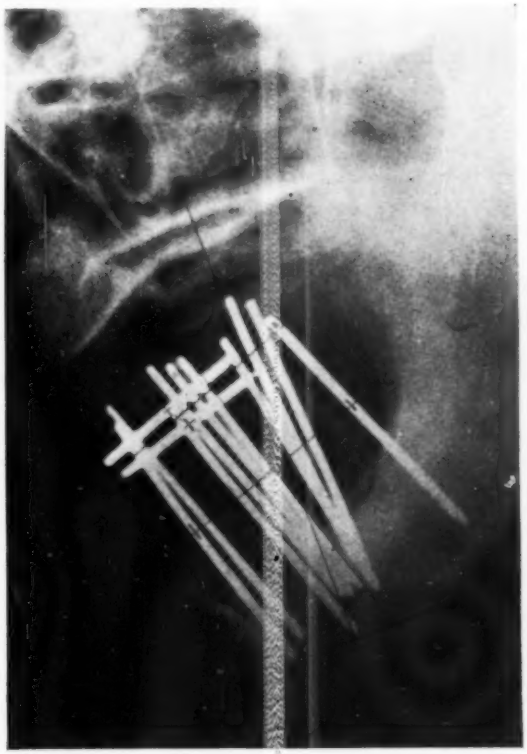


FIG. 304.—Volume type of needle implant.

untreatable by any other means. Moreover, in the event of failure, cases previously possible for surgery have a second chance by this method.

TOTAL RESULTS

That the advantages of radium are indeed combined with an ability to produce useful results is illustrated in *Table I*, which is an addition of the figures published in our three statistical reports. A reminder about staging: Stages I and II consist of cases with lesions up to 4 cm. in diameter, but without secondary nodes; Stage III consists of primary lesions up to the same extent, but presenting with mobile secondary nodes; Stage IV contains the cases more advanced. For the whole group the five-year figure is 32 per cent, and I find that the best figure we have published for the floor of

the mouth is 58 per cent for 39 Stage I cases in the 1934–8 report.

Group for Analysis.—The above group dates from 1932 when radium techniques were not very well established—I thought it would be more representative to take for analysis a later group covering the eleven years 1935–45, and consisting of 345 treated cases.

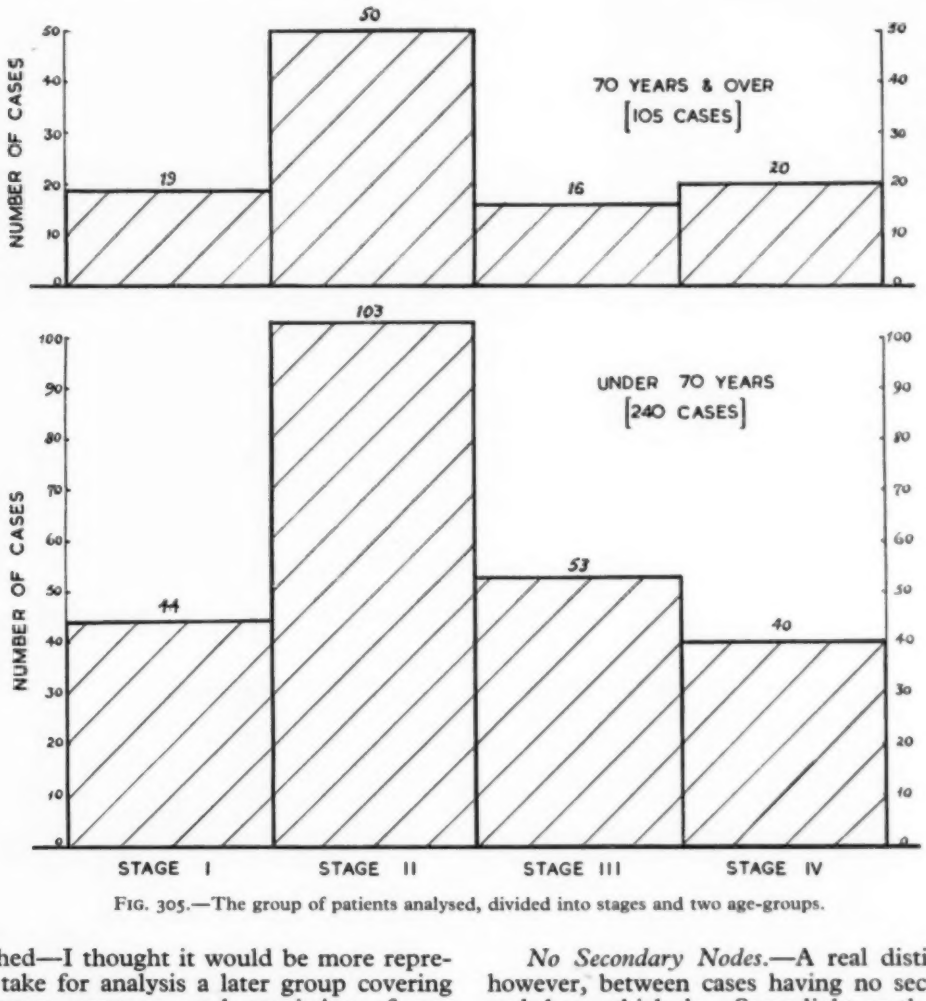


FIG. 305.—The group of patients analysed, divided into stages and two age-groups.

Table I.—CARCINOMA OF FLOOR OF MOUTH—RADIUM—1932–44
(*Christie Hospital and Holt Radium Institute, Manchester*)
Summated Results from Three Published Statistical Reports

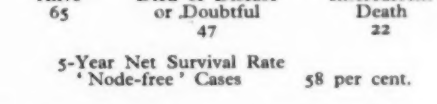
STAGE	NUMBER TREATED	5-YEAR NET SURVIVAL RATE
		Per cent
Early (I/II)	247	43
Late (III/IV)	180	18
Total	427	32

The composition of this group is shown in *Fig.* 305. The proportion of patients aged 70 and over amounts to 30 per cent, and the more treatable cases, meaning by that Stages I, II, and III under 70, make up 58 per cent.

On looking through the records, no great contrast is found in the primary lesions in Stages I, II, and III—eight diagrams picked at random from the Stage I and II groups are reproduced in *Fig.* 306.

No Secondary Nodes.—A real distinction exists, however, between cases having no secondary nodes and those which do. Some light on the effectiveness of radium treatment is afforded by analysing Stages I and II, no-node cases, as shown in *Table II.* Sixty-two per cent of them remained free from secondary nodes, and 58 per cent of these survived for five or more years. This figure happens to coincide with the figures already given for a small group of Stage I cases.

Table II.—CARCINOMA OF FLOOR OF MOUTH—RADIUM—1935–45. STAGES I/II ONLY
(*Christie Hospital and Holt Radium Institute, Manchester*)



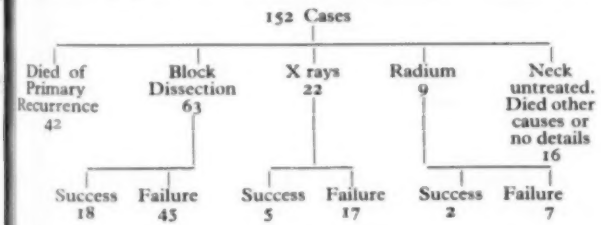
5-Year Net Survival Rate
'Node-free' Cases 58 per cent.

The other observation that one draws from this table is that about two-thirds of the patients were never in need of treatment for secondary nodes—an important consideration in the case for or against prophylactic block dissection.

Prophylactic dissection has, in fact, not been the practice, frequent examination being preferred, with block dissection as soon as nodes can be found by the fingers.

With Secondary Nodes.—The effect of mobile secondary nodes, present or later appearing, is analysed in *Table III*, which consists of all cases originally Stage III, plus the 82 cases from *Table II*, and refers to the treatment of the nodes only.

Table III.—CARCINOMA OF FLOOR OF MOUTH—RADIUM—1935-45. NODES PRESENT OR DEVELOPED LATER. (STAGE IV EXCLUDED)
(*Christie Hospital and Holt Radium Institute, Manchester*)



In this group a rather disappointing fraction qualified for block dissection, which has always been regarded as the method of choice, and the history of these patients is summarized in *Table IV*. Among the failures there are 19 primary recurrences, but 30 recurrences in the neck.

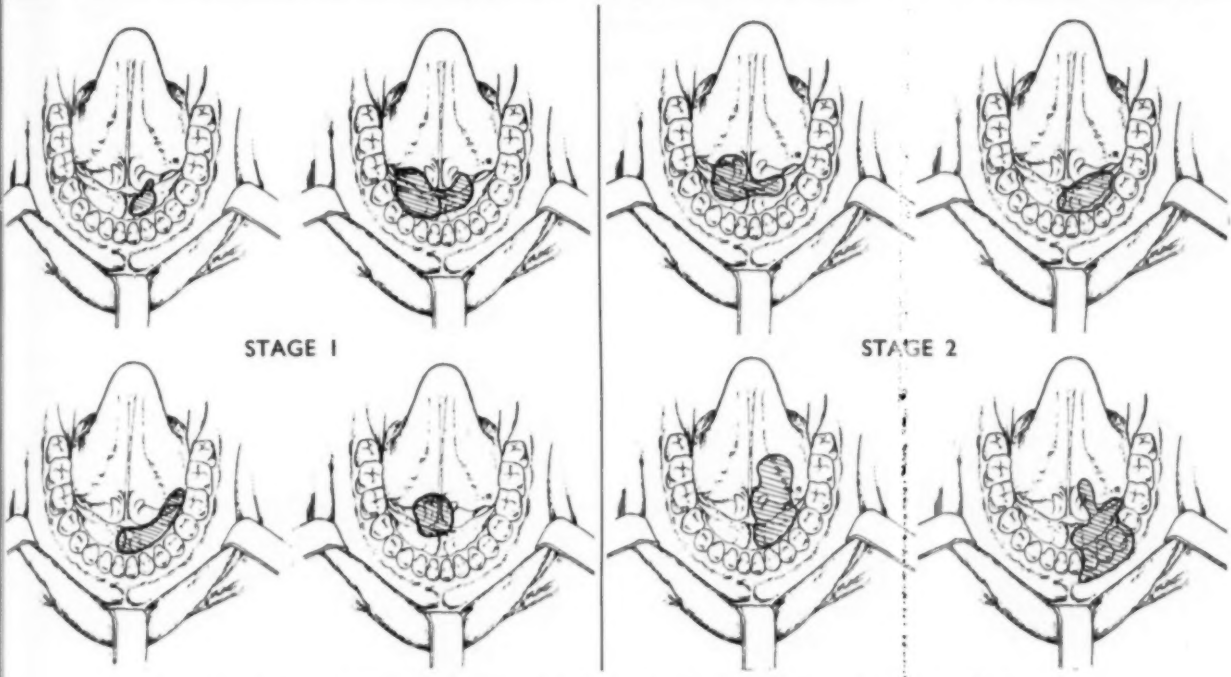
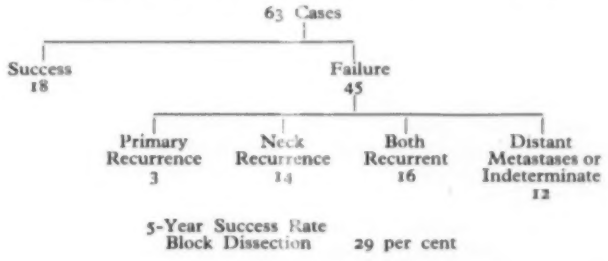


FIG. 306.—A group of diagrams taken at random from patients' records, to show size and site of lesions.

Table IV.—CARCINOMA OF FLOOR OF MOUTH—RADIUM—1935-45. BLOCK DISSECTION GROUP
(*Christie Hospital and Holt Radium Institute, Manchester*)



CONCLUSIONS

The evidence, as a whole, may be conveniently summarized as showing a cure-rate of about 60 per cent for early cases and for cases which do not develop secondary nodes, and 30 per cent for cases qualifying for block dissection, and also for the whole group of patients, including all ages and stages.

COMPLEX FRACTURES OF THE MIDDLE THIRD OF THE FACE AND THEIR EARLY TREATMENT

By R. L. G. DAWSON and G. L. FORDYCE

FROM THE PLASTIC AND JAW UNIT, HILL END HOSPITAL, ST. ALBANS

It is unfortunate that in English there is no comparable term to the *machoire supérieure* of the French which incorporates the thirteen bones of the middle third of the face.

In the following account of injuries to this area, for lack of better terminology, the name ' fractured maxilla ' will be used to describe bone injuries to the central middle third of the face, excluding separate local injuries to the nose. The fractured malar-maxillary-zygomatic complex, apart from local fractures of the zygomatic arch, will be called the ' fractured malar '.

This paper is based on a review of 190 cases of fractured maxillæ received in civilian accidents.

A fracture of the maxilla is caused by a direct blow on the face. The direction of the displacement, the severity of the fracture, and the presence or absence of fractures of other bones of the face depend upon the force and the direction of the blow. It is indeed fortunate that there is, anterior to the cranium itself, a mass of cellular bone that can absorb a large amount of an injuring force before the effect of the blow falls upon the cranium, for without such a buffer there would assuredly be many more fatal head injuries.

Nevertheless, one should not be content that the injured is fortunate to have escaped with so little cerebral trauma, and disregard the deformed area that has saved his life.

Other facial injuries, such as a fractured mandible or malar bone, lacerations, fractures of the frontal bones and base of the skull, and the results of trauma to the rest of the body, often complicate the diagnosis, management, and prognosis of the fractured maxilla (*Tables I, II*).

In the treatment of these injuries *function* must be restored to the face and this necessitates the replacement of the tooth-bearing areas into normal

Fig. 307.—An unreduced case. Note the pseudo-hypertelorism, flat nasal bridge, flat right malar bone with a lower position of the right eye, a retroposed maxilla, and almond-shaped palpebral fissure.

occlusion with the mandibular teeth, reduction of nasal fractures, and straightening the deformed nasal septum to ensure a clear airway. Associated depressions of the malar must be corrected to bring the eye on the affected side up to its original level, to reduce the possibility of a permanent diplopia, and to allow a free movement of the coronoid process of the mandible within the zygomatic arch.

Table I.—Associated Facial Bone Injuries Superimposed on the 190 Fractured Maxillæ

	Cases
Fractured malar-zygomatic compound, displaced or not displaced	125
Fractured displaced nasal bones	106
Fractures of the mandible, of all types	85
Fractured, splayed, ethmoid area	32

The ultimate *appearance* of the patient will depend upon how well the bones have been replaced into their normal position, avoiding the frightful ' dish-face ' appearance with the retroposed maxilla, flattened nose, the pseudo-hypertelorism of the splayed-out ethmoid bones, and the psychological and social upsets associated with these deformities (*Fig.* 307).

It will also be shown later that accurate replacement and immobilization of the displaced skeleton

is the greatest factor in controlling the cerebrospinal rhinorrhœa, often associated with these fractures.

Table II.—Associated General Injuries

	Cases
Involvement of the central nervous systems including concussion	47
Fractured skull	36
Fractures of the lower limbs	14
Fractures of the upper limbs	33
Lacerations of the trunk and limbs	13
Severe eye injuries	6
Burns from exhaust pipes and car fires	4
Joint injuries of the limbs	2

Early replacement is also essential to avoid the pharyngeal obstruction that occasionally occurs in a severe injury of this type.

THE INCIDENCE

Table III lists the percentage incidence in the 190 cases analysed.

Table III.—The Percentage Incidence of Types of Accident in 190 Cases

	Per cent
Motor-cyclists	32
Motor car occupants	23
Pedal cyclists	16
Pedestrians hit by vehicle	7
Industrial and aeroplane accidents	22

CLASSIFICATION

In 1900–1901 Le Fort, whose classification was introduced into this country by James and Fickling (1941), determined the salient fracture lines by a series of experiments in which he applied force of varying direction and strength to cadaver heads.

McIndoe's (1941) classification covers too wide a field, in that he includes simple fractures under the same heading as complex injuries.

Ivy and Curtis (1938), on the other hand, introduced a classification which is on the whole too simplified to be of value. It is of interest that the fractures under review follow the fracture lines as described by Le Fort fifty years ago. The classification of these fractures is Le Fort's (*Table IV*).

Table IV.—The Percentage Incidence of Types of Fractured Maxillæ, based on Le Fort's Classification

	Per cent
Unilateral Le Fort I	10
Le Fort I	23
Le Fort I/II	21
Le Fort II	34
Le Fort II/III	2
Le Fort III	2
Le Fort I/III	4
Le Fort I/II/III	2
Unilateral Le Fort II	1
Le Fort I/Unilateral Le Fort II	1

Type I.—The fracture line passes horizontally above the alveolus and palate, through all walls of the maxillary antra, across the nasal septum, and low down through the pterygoid processes (*Figs.* 308, 309).

Type I. Unilateral.—The maxilla is fractured through the same lines as Type I but is split down the centre of the palate and does not extend to the opposite side. It is often associated with a fractured displaced malar on the same side, and is caused by a lateral blow.

Type II.—The fracture lines pass across the nasal bones and the frontonasal processes at varying heights, while on each side they cross the lacrimal bone, doubling back through the orbital plate of the maxilla and the infra-orbital margin. They then descend laterally through the anterior wall of the antrum, round the malar-maxillary buttress, and across the posterior wall of the antrum, into the pterygo-maxillary fissure, finally fracturing the pterygoid processes. The fracture is often associated with a fractured displaced nose and flat splayed-out ethmoid bones (*Fig.* 308).

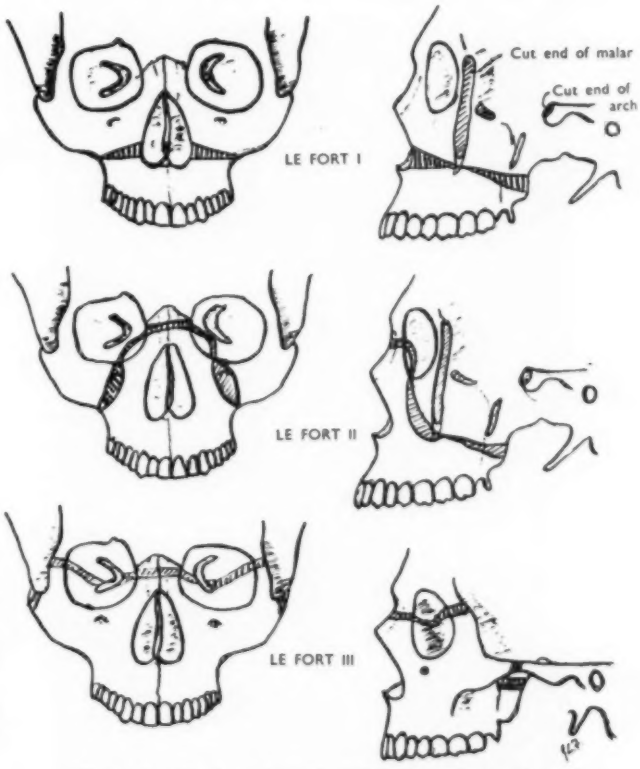


Fig. 308.—To show the region of the fracture lines in each of the three main types. Note that the pterygoid plates are broken in each type. The fracture line is indicated by dense hatching.

Type I/II.—Is a combination of both these separate types, and as the nasomaxillary complex is separate from the alveolar and palatal element, two different degrees of displacement may occur.

Type III.—Is a complete craniofacial dysjunction. The condition is very rare. The horizontal fracture line passes across the frontal process of the malar on each side, medially across the back of the orbit, through the inferior orbital fissure, below the optic nerve, and across the sphenoid, ethmoid, and lacrimal bones and the frontonasal processes of the maxilla. It passes across the junction between the nasal bones and the frontal bone and fractures the nasal septum high up. It also fractures the upper part of the pterygoid processes and the zygomatic arch on each side. The facial bones, therefore, have become separated as a whole from the base of the skull (*Fig.* 308).

It must be understood, however, that the fracture lines described above are only the prominent

ones, and that the antral walls, pterygoid plates, the septum, and paper-thin bony structures are comminuted to a degree proportional to the blow received. The single fracture line of the classification courses through these areas of bone comminution indicating the level of the damage. *Fig.* 309 shows clearly this comminution.

The antral wall may be stove-in, the fractured malar-maxillary buttress may impact deep to the malar bone, or the fractures of the posterior antral wall may overlap.

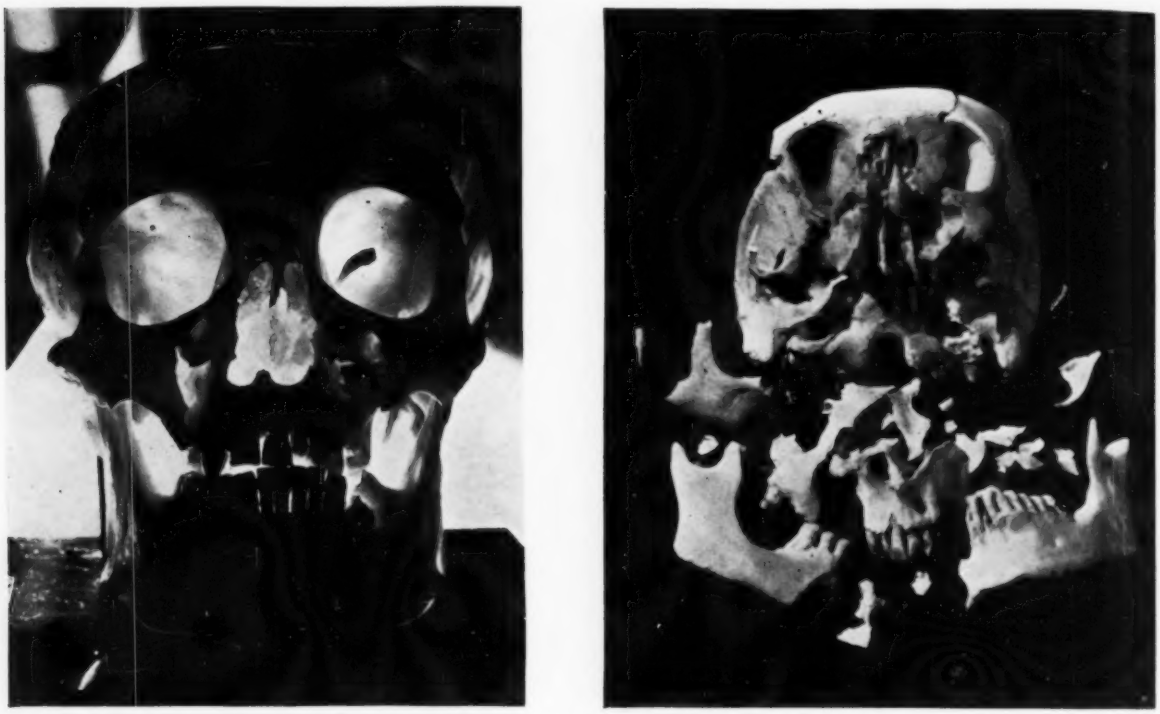


FIG. 309.—Two disarticulated specimens of facial fractures. The specimen on the left has been re-assembled. It is a Le Fort I type. Note the deep retroposition of the lower part of the maxilla and comminution of the antral walls. The specimen on the right is a Le Fort II type with an associated fractured right malar, frontal bone, and mandible. The multiplicity of the fragments of this typical case should be noted.

The nasal septum is, except in unilateral cases, always fractured. The most severe septal lesions, with comminution and overlap of the fragments, occur with a direct anterior blow to the nasal bridge. Generally, the site of fracture of the septum corresponds with the type of maxillary fracture. It is low in Type I and high in Type III.

In addition to various degrees of fracture of the antral walls, pterygoid plates, and the nasal septum, the lacrimal bones, the lamina papyracea of the ethmoids, the walls of the ethmoid air-cells, the cribriform plate, and the orbital plate of the frontal bone may be involved to a greater or less degree.

These numerous and irregular fragments are occasionally a source of difficulty when reduction is attempted.

THE MECHANISM OF THE DISPLACEMENT

The displacement of the fractured maxilla is mainly determined by the force and direction of the blow. The maxillary block may be impacted upwards and backwards, or may be free, and therefore termed 'floating'. Lateral displacement is common. Gagging of the bite in the molar region due to downward displacement posteriorly is a frequent finding.

Fracture of the bony origins of the medial and lateral pterygoid muscles throws them into spasm, which results in a downward and backward pull on the separated fragment and accounts for the trismus often encountered. This displacing force is enhanced by the action of the superior constrictor of the pharynx, and by the palato-pharyngeus and palatoglossus muscles, which have origin on the fragment (Bataille, 1947). In cases of floating maxillæ the action of these muscles may be observed when during deglutition the maxillary block is seen to move.

These factors and the effect of gravity make it imperative that immobilization be applied following reduction and make it obvious that, save in very exceptional circumstances, reduction without fixation is doomed to failure.

The degree of displacement and comminution of the nasomaxillary complex is of the utmost importance as this is the greatest factor governing the ultimate æsthetic result.

Simple fractures of the nasal superstructure are readily appreciated, and treated, but in the more severe case the nasal bones, frontonasal processes, and the lacrimal bones are pushed back as a comminuted mass into the anterior ethmoidal air sinuses. At the same time the nasal contour is flattened out to produce an unsightly pseudo-hypertelorism. This

mass of highly comminuted, thin bony particles quickly organizes itself, with the aid of a large amount of ragged periosteum and blood-clot, into a solid mass, which if it is not reduced within five to seven days, will become irreducible, except by an open operation. To distinguish this fracture from other nasal fractures we call it the ' fracture of the ethmoids '.

It should be emphasized, again, that immobilization is necessary to maintain the reduction of the displacement.

A B

FIG. 310.—A, A case on admission. Note the bilateral orbital ecchymoses, the wide nasal bridge, and the grossly swollen face. B, This man is attempting to close his teeth. Note the gagging present in the left molar region, as shown by the anterior open bite, also the difference in level between the upper incisors due to a central split of the palate, buccal sulcus ecchymosis, and retroposition of the maxilla.

SYMPTOMS

Compared with the severity of the lesion the symptoms are few. Epistaxis will have occurred, and marked tightness of the face with swelling and difficulty in opening the eyes through œdema, and hæmatoma is present. Anosmia, and a constant discharge of cerebrospinal fluid down the nostrils or into the pharynx, may be found in the high fractures of Le Fort II and III types. Nasal deformities may be noticed by the patient.

There may be some difficulty in swallowing and breathing on account of mechanical obstruction by hæmatoma or œdema of the soft palate, or from the severe degree of downward and backward displacement of the maxilla, which causes the palate to rest on the dorsum of the tongue. The presence of blood-clots in the pharynx may also cause great difficulty with breathing.

The fractured maxilla itself may cause some ill-defined pain on movement, felt around the area of the upper buccal sulcus. Pain is felt on attempting to bite, and failure of the upper and lower teeth to occlude together may be noticed by the patient, but in our experience statements about the previous occlusion are often fallacious.

Infra-orbital anæsthesia, through damage to the maxillary nerve in its canal, may occur in cases of Le Fort II fracture or where there is a fractured depressed malar bone. In the latter fracture diplopia can be an additional symptom. When the zygomatic arch is depressed difficulty in mouth opening may be encountered, while in coincident fracture of the mandible, pain, intra-oral bleeding, a deranged bite, and local swelling are common symptoms.

SIGNS

Once having learnt the cardinal physical signs of the fractured maxilla, such injuries can be diagnosed within a few minutes.

The patient is rarely shocked, for the actual mass of tissue damage is small. Unless severe hæmorrhage or prolonged respiratory obstruction has occurred, the presence of shock indicates other severe lesions.

The patient presents a very typical appearance, with symmetrical swelling of the face, bilateral orbital hæmatomata, subconjunctival hæmorrhages, and swollen eyelids (*Fig.* 310 A). When there is marked retroposition of the maxilla the face appears flat in spite of the swelling, and may look longer or shorter than normal. An associated depressed malar may be palpated, and confirmed by the lower level of the eye on the affected side.

The infra-orbital margins will be irregular in the Le Fort II type, indicating the site of fracture,

and a lateral tilt of the maxilla may be diagnosed from the level of these fractured surfaces, which can be felt on manipulating the maxilla. The Le Fort I and III types have intact infra-orbital margins, while in the Le Fort III type pain may be elicited over the

FIG. 311.—A lateral radiograph of the face to show the fracture of the pterygoid plates.

external angular processes and the zygomatic arch on each side.

Sensation may be absent in the area supplied by the infra-orbital division of the fifth nerve if the malar is fractured, or if the Le Fort II fracture lines have passed through the infra-orbital canal.

The intra-oral displacement of the maxilla is often found to be backward, with a tilt downward in the molar region, and a lateral shift may be observed on attempting occlusion with the mandibular teeth (*Fig.* 310 B). The maxilla can be firmly impacted or floating. The presence of bilateral, upper buccal sulcus ecchymoses, and palpation of the fractured malar-maxillary buttresses confirm the diagnosis.

A central palatal irregularity indicates a fracture of the hard palate. A hæmatoma of the soft palate may occur in any fracture of the maxilla. Some degree of trismus is a constant finding.

Tapping the teeth with a metal instrument will give a ' cracked pot ' note if the maxilla is fractured, which is itself diagnostic. Although readily elicited by the dental surgeon, this test may not be so useful to the surgeon unused to assessing the percussion notes of teeth. A development of this is the tuning-fork method, first used by one of us (G. L. F.), which is useful on the conscious and rational patient. The vibrating tuning-fork is held on the forehead and on the maxillary teeth. On the intact maxilla the tooth note is much louder, to the extent of being unpleasant, than on the forehead, whereas on the fractured maxilla the note is dulled.

The dental surgeon should be consulted with reference to the bite in the hope that a study of the teeth and their facets will indicate the pre-accident occlusion. If there is any doubt about this, plaster models of the teeth should be prepared and occlusion tested. In the edentulous patient, his dentures, if available, should be tried in the mouth to indicate the displacement.

Examination of the nose will reveal either separately displaced nasal bones, a torn and broken septum, a septal hæmatoma, or nasal bones moving with the maxilla. Where there is ethmoid bone involvement there is a marked widening, both visible and palpable, between the inner canthi (*Fig.* 307). Cerebrospinal fluid may flow down the nose or into the pharynx, in which case the patient will often state that fluid is coming down the back of his throat, and he is seen to be swallowing frequently. Jugular compression will increase the flow and confirm the diagnosis.

Externally the frequently associated fractured mandible may show swelling and deformity, whilst

FIG. 312.—A 30° occipitomental view of a Le Fort I type to show the fracture of the malar buttress on each side. Note the presence of bilateral opaque maxillary antra.

intra-orally dental irregularity and a torn, bleeding mucosa can often be seen. An associated fracture of the mandibular condyle will cause swelling and tenderness. Condylar movement on the affected side may be reduced or even impalpable. Malocclusion consequent upon such a fracture will materially complicate the assessment of the maxillary displacement.

RADIOLOGICAL INVESTIGATION

The taking of numerous radiographs of the face is not only a tiresome procedure for the injured person, but is often in fact relatively unnecessary.

FIG. 313.—A 30° occipitomental view of a Le Fort II type. Note fractures of both orbital floors, with impaction downward on the left side, and fracture of the malar buttresses and high across the nasal bridge. Also note the fractured septum.

There are very few cases in which X-ray films are essential to diagnosis, and nearly every case *can* be diagnosed and treated satisfactorily without their aid. Radiography is useful to demonstrate a coincident fractured skull, fractured frontal sinuses, a comminuted malar, and associated minor ramifications of the fracture lines which are hidden when the face is enormously distorted by swelling, and when consequently good palpation is difficult. It is also used to determine whether unaccounted teeth are in the antra or the chest. Radiographs provide a record of the pre-reduction state and are often required in litigation cases.

Variations of the occipito-mental view for the sinuses are taken. These, however, show such a mass of bone lines superimposed and crossing each other that close examination is confusing, and a general view at a distance suffices to give the necessary information. In the lateral skull view the palate may be seen to be retroposed, a fracture line passing just above it in the Le Fort I type, while the corresponding fracture line in a Le Fort II and III will be seen crossing the frontonasal processes and the nasal bones, sometimes passing backwards into the ethmoid region. The pterygoid processes and the posterior wall of the antrum will invariably show a break (*Fig.* 311). In the 30° and 60° occipitomental views, the transverse fracture lines of the three principle types show up well. The low transverse lines of the Le Fort I (*Fig.* 312), the fractured infra-orbital margins and fractured nose of the Le Fort II (*Fig.* 313), and the fractured posterior orbital wall of the Le Fort III (*Fig.* 314), should be looked for. Bilateral opaque antra will confirm a maxillary fracture unless enormous facial swelling is present, which itself gives an impression of clouded antra. A coincident injury to the malar bones can be seen.

DIFFERENTIAL DIAGNOSIS

Although a fractured malar, fractured nose, fractured frontal bones, and a stove-in nasal bridge and ethmoid bones may coexist with a maxillary injury, a firm maxilla, in normal occlusion with the mandible, will rule out the latter.

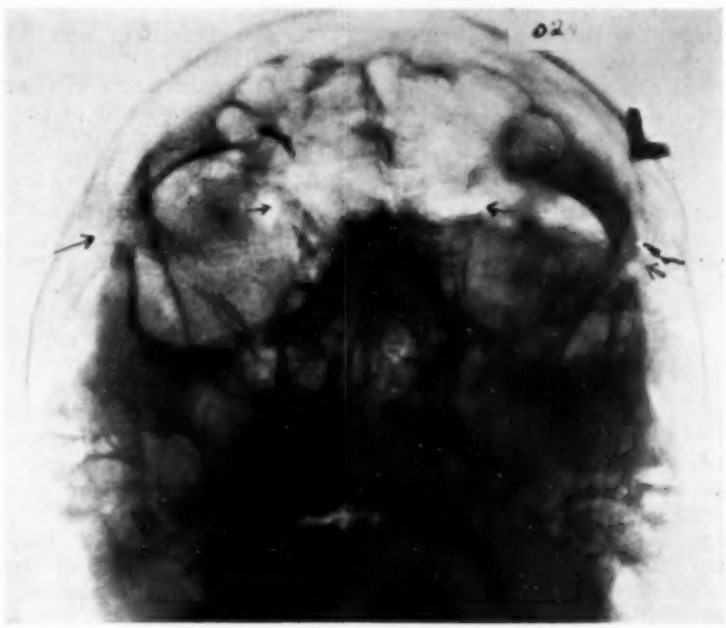


FIG. 314.—A 30° occipitomental view of a Le Fort III type with intact orbital floors, but displacement of the external angular processes, and gross comminution across the nasal bridge, and the lower part of the frontal sinuses.

A recently *fractured malar bone* may have any or all of the following signs: Unilateral orbital hæmatoma; inferolateral subconjunctival hæmorrhage; depression of the orbital margin, and malar prominence; anæsthesia of the cheek, upper gingivæ, and teeth; unilateral epistaxis; and occasionally diplopia and enophthalmos.

A B

FIG. 315.—A, Introducing Walsham's forceps to rock the maxilla and disimpact it. Note rubber covering for the palatal arm of the forceps. B, Forceps in place.

A *depressed zygomatic arch* will in addition to the palpable deformity, sometimes show interference with mouth-opening, because the coronoid process of the mandible impinges on the depressed bony fragment of the arch, or because of spasm in the masseter muscle.

Swelling and displacement of the *nose* and *septum* alone points solely to a nasal injury. When this is combined with a widening in the canthal region it indicates the more severe, *stove-in injury* of the nasal bridge. An additional confirmatory sign of this injury, if the lid swelling is not great, is the alteration of the palpebral fissures from the normal ellipsoid to an almond shape, due to lateral displacement of the medial palpebral ligaments.

A fracture of the *frontal bone* may show bilateral orbital hæmatomata and subconjunctival hæmorrhages, but there is usually a local swelling over the site of the injury, and the hæmorrhage and eyelid swelling appears first in the upper half of the orbit.

TREATMENT

The *first-aid treatment* is to secure a clear airway and to stop any severe hæmorrhage arising from facial wounds or intra-oral lacerations. The mouth should be cleaned out, any loose teeth and dentures removed, and the patient placed in the prone position, with the tongue pulled forward. If the maxilla is pushed back a long way, the soft palate occludes the nasopharynx, and the dorsum of the tongue rests against the hard palate. This will obstruct the airway and the maxilla should, therefore, be pulled forward with a finger hooked around the palate. A nasopharyngeal tube should be introduced if there is any question of inadequate ventilation. Unless there is a serious injury to the larynx, tracheotomy is not necessary. It is imperative that pharyngeal toilet, adequate positioning, and the introduction of a nasopharyngeal tube be performed *before transfer by ambulance*. If these first-aid principles are followed, the patient will travel well; but they have, on occasions, been admitted *in extremis* when they have not received adequate first aid prior to their journey.

The *routine* of the *definitive treatment* of these injuries is an orderly sequence and should be:—

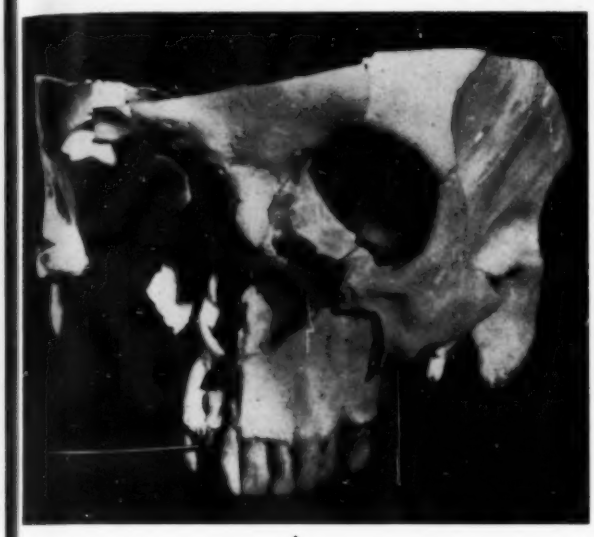
1. Any necessary mobilization of impacted fractures.
2. The reduction and fixation of the tooth-bearing fragments.
3. The reduction and fixation of the lateral and nasomaxillary elements of the injury.
4. The repair of intra- and extra-oral lacerations.
5. The removal of the pharyngeal pack and the application of intermaxillary fixation.

The closest co-operation between surgeon and dental surgeon is essential during any operative procedure. In a small proportion of cases no operative treatment is indicated. An edentulous patient, showing minimal displacement of the maxilla, and with no appreciable change in the facial contour, may be provided with a satisfactory compensation for any jaw irregularity by suitable dentures. Likewise, a patient showing merely a crack fracture without disruption of the normal occlusion, may be treated expectantly. When operative procedures are indicated, general anæsthesia should be used.

1. Mobilization of Impacted Fractures.—The essential principle of reduction is to mobilize fully any impacted fragments. Associated depressed malar components must often be elevated before the maxillary segment can be brought forward. The maxilla is then reduced either manually or with Walsham's forceps, until a proper occlusion with the mandibular teeth is obtained. Often the maxilla is sufficiently impacted to need rocking free with the forceps, gripping the palate on both nasal and palatal

aspects (*Fig.* 315). Special difficulty may be met, when reduction is attempted, from impaction of the maxillary fragments beneath the remainder of the stable facial skeleton.

In some cases of backward impaction the medial margins of the malars prevent forward reduction. The malars must then be sprung outwards with



A

B

C

FIG. 316.—A, Actual post-mortem specimen to demonstrate impaction deep to the malar. B, To show how the fronto-nasal processes may be separate from the tooth-bearing fragment. C, To demonstrate impaction of the frontonasal processes deep to the infra-orbital margin.

Bristow's elevators inserted beneath the zygomatic arch, through bilateral buccal incisions, while simultaneous forward traction is made on the maxilla.

An impaction of the maxilla deep to the malar on one side will require disimpaction by increasing the malocclusion, pulling the whole maxilla to the opposite side, and finally bringing it to the central position (*Fig.* 316 A). A separate small fragment may impact between the malar-maxillary buttress and the malar bone, and removal of this fragment through a buccal incision will then be necessary to achieve reduction. Where the posterior antral wall has been fractured, and the inferior fracture line has impacted behind the superior fragment, the maxilla will have to be brought downward at the back before pulling it forward.

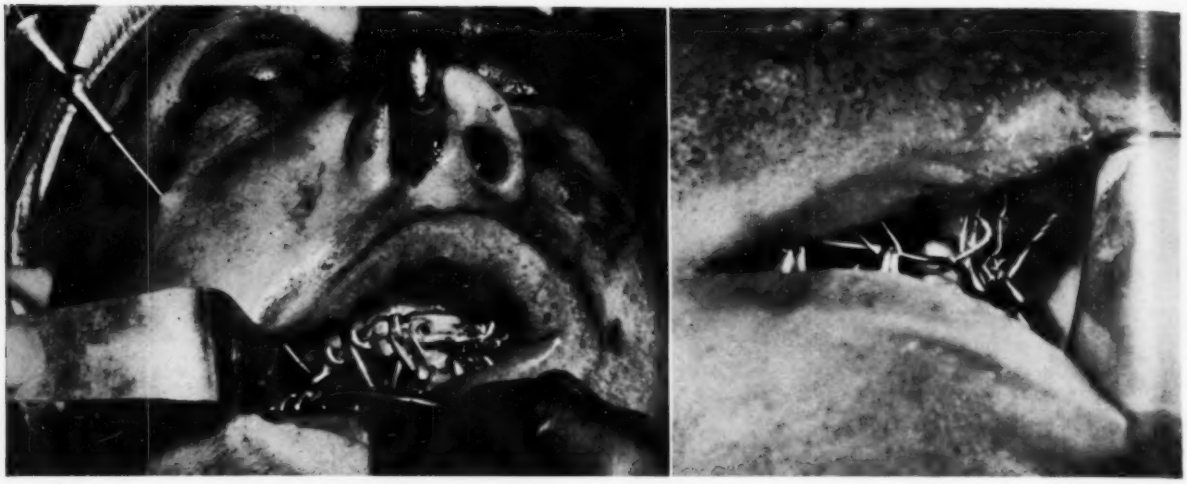
The medial fragment of the infra-orbital margin of the Le Fort II type fracture may impact beneath the stable lateral portion (*Fig.* 316 C). This may require open reduction through a small incision along the line of the infra-orbital margin. If the incision is large, œdema of the lower eyelid lasting for some months will follow owing to lymphatic obstruction.

Lastly, in a small minority of fractures, especially of the combined Le Fort I/II types, the central nasomaxillary complex fails to come forward when the tooth-bearing segment is reduced (*Fig.* 316 B). Levers must therefore be passed up behind the frontonasal processes through bilateral antrostomy incisions, to disimpact the central block and allow it to be brought forward.

When all impacted fragments have been mobilized, reduction is possible, and the problem of fixation arises.

2. Reduction and Fixation of the Tooth-bearing Fragments.—Prior to anæsthesia a plaster headcap is applied, over stockinette, taking care to get the plaster below the occipital protuberance, and over the vertex (Mowlem, 1941). A German silver frame is incorporated and projects from the frontal region of the headcap. Dental cap splints are cemented to the upper and lower teeth (*Fig.* 317 A). Basically the fixation may be applied in two ways, either directly from the headcap (craniomaxillary) or sandwiched between the headcap and the lower jaw (craniomandibular). We prefer, where possible, to use a craniomaxillary fixation, which has the advantage that intermaxillary fixation is not an essential

part of the procedure and the mouth can, therefore, be opened. This is not only much more pleasant for the patient, but allows better pulmonary ventilation part, allows the patient to masticate a normal diet, and permits access to intra-oral lacerations. Oral hygiene can in many cases be carried out by the



A B

FIG. 317.—A, Cap splints applied. Intermaxillary elastics on. Passing a cheek wire via a lumbar puncture needle. B, Jelenko arch wires in use. Note the cheek wire on extreme right of mouth passing to the mandibular arch wire.

in those cases which suffer from an embarrassing degree of nasal obstruction or have pulmonary complications.

A serious disadvantage of the craniomandibular fixation is that the mandible may be secured to the patient himself, relieving the nursing staff of an arduous and tedious task.

Craniomandibular fixation presents a technical difficulty in that in cases where a good complement of teeth are present removal of the throat pack at

A B

FIG. 318.—A, On admission, showing bilateral orbital ecchymoses, and a flat, wide, and deformed nose. B, The same case, lateral view, to show the retroposition of the centre of the face.

head cap in an eccentric position which may not be appreciated until the patient recovers from the anæsthetic and complains of temporomandibular joint pain, or, where there is minimal discrepancy, until the fixation is removed.

Craniomaxillary fixation with free movement of the mandible encourages the blood-supply to the the termination of the operation necessitates temporary undoing of the cranial support, thus jeopardizing the reduction already achieved.

Where no dental laboratory facilities are available, arch wires of the Jelenko or Winter pattern, fixed to the teeth with stainless steel wire, and suspended from a headcap, are very useful, but have the

disadvantage that the fixation is craniomandibular (*Fig.* 317 B).

A stainless steel wire is passed through the cheek on each side in front of the malar prominence, by way of a standard lumbar puncture needle (Mowlem, 1941; Federspiel, 1934). The entry site of the cheek wire is so placed that the wire will pass in a straight line from the headcap to the maxillary splint and approximates to the point of least movement in the cheek, thereby avoiding subsequent cutting of the skin by the wire (*Fig.* 317 A). These wires are attached to loops on the cap splints in the first molar region, and to the lateral sides of the frontal horns. A screw-traction apparatus is incorporated with the wires so that they may be tightened or loosened at will.

The maxilla, having been suspended laterally and posteriorly by the cheek wires, is now fixed in front by a steel bar from the headcap to an anterior bar from the upper cap splint.

There now being both a fixed upper limit, i.e., the headcap, and a fixed lower limit, i.e., the maxilla, the further fragments of the fractured face can be fitted into place.

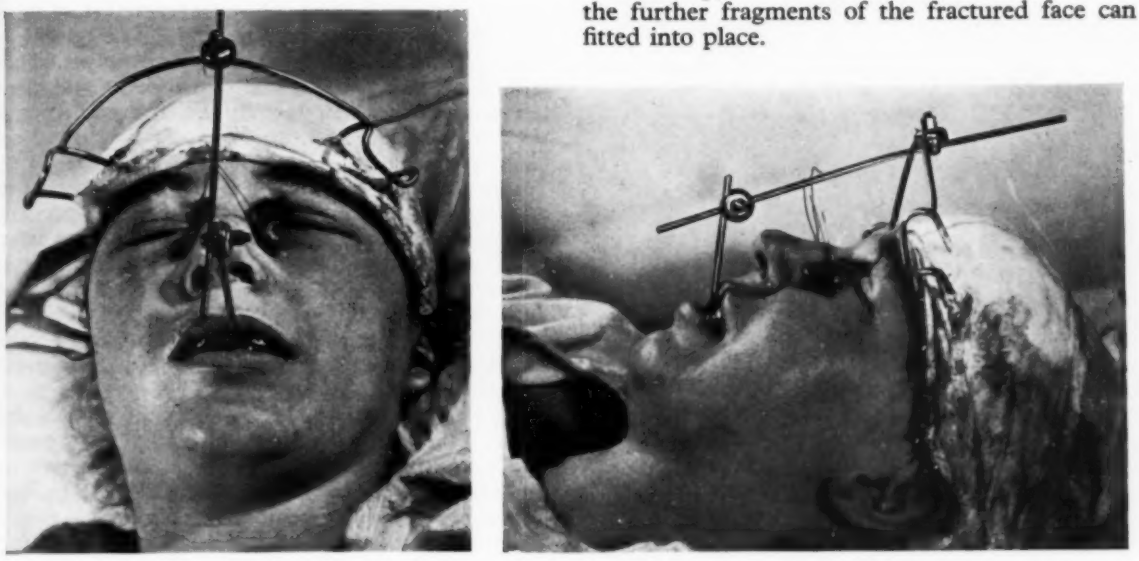


Fig. 319.—Craniomandibular fixation has been used for a split palate was present. Note the position of the nose, and compare it with *Fig.* 318 B. The lateral nasal plates with transnasal fixation is shown.

3. The Reduction and Fixation of the Lateral and Nasomaxillary Elements.—

The *nose*, if fractured separately, is disimpacted, any septal trauma repaired where possible, and a plaster fixation applied. If the nose has been pushed backwards, with the nasal bones splayed laterally and the septum buckled, then fixation is maintained by wire or thick silk passed through both sides of the nose behind the nasal bones, and tied over lead plates or cotton-wool rolls, on each side. This fixation corrects the lateral spread of the nose.

FIG. 320.—The same case after removal of all fixation. The nose and face are now forward, but the space between the inner canthi is too wide.

For those cases with crushed, splayed-out, fractured 'ethmoid bones', and a widening of the nasal bridge at the level of the inner canthi, the nose is elevated with Ash's septal forceps at the same time as the ethmoids are pinched together between the

FIG. 321.—To show, on the patient's left, the method of screw-pin fixation for a fractured malar. Also shows bilateral nasal plates to correct a depressed nasal bridge.

finger and thumb, but the operator should not consider a good reduction has been obtained until it appears as if the finger and thumb are practically meeting behind the frontonasal processes. The position is maintained by silk or wire passed on a large curved, cutting needle through the fracture lines at the root of the nose as close to the medial palpebral ligament as possible. The aim of fixation close to the attachment of the medial palpebral ligament is to maintain reduction of the bone fragment to which the ligament is attached. It is the lateral displacement of this fragment, which may be primary, caused by the displacing force, or secondary, due to action of the orbicularis oculi, that causes the unsightly almond-shaped palpebral fissure. Fixation must be applied, as the action of the orbicularis oculi will cause re-displacement (*Fig.* 319).

The reduction of the ethmoid block is the most urgent procedure in the repair, and a delay of four or five days will prevent full reduction being obtained. With the central masomaxillary complex pushed back and separate from the alveolus and palate it is necessary to lever the frontonasal processes forward via bilateral antrostomies. The complex may then be held forward with a wire passed behind the frontonasal processes and attached to the frontal bar, *in conjunction with* lateral compression of the nasal elements (*Figs.* 318–320).

The nasal fixation is maintained for ten to fourteen days, and following removal a probe is passed along both sides of the septum to ensure that no synechiæ exist.

The *fractured and displaced malar* is elevated through a temporal approach above the hairline. If the fracture is of a simple type the bone will maintain its position with no fixation. If the malar has been comminuted or if there is a central depression or comminution of the orbital floor, the antrum should be opened through the upper buccal sulcus and the fragments reduced digitally. If the fragments are unstable the antrum should then be packed with ribbon gauze soaked in acriflavine and paraffin emulsion or penicillin cream. The packing is removed without anæsthesia in two stages at seven and twelve days. If the main bone fragment is unstable good contour may be restored by fixing the bone to the headcap by a stainless steel screw (*Fig.* 321).

Direct bone wiring of the fracture at the external angular process is occasionally a useful procedure, especially when the rest of the malar bone is comminuted and an antral pack is required. Stabilization, with one loop of 35-gauge stainless steel wire, prevents over-correction when the pack is inserted, while ensuring that the lateral canthus is correctly reduced and therefore the palpebral fissure in proper alinement with the opposite side.

A warning must be given regarding the possibility of displacing the maxillary block laterally by overpacking the antrum, and therefore such a pack *must* be introduced after immobilization of the tooth-bearing segment has been effected.

For *associated fractured mandible* the appropriate reduction and fixation is performed. A fractured mandible, however, complicates the reduction of the maxilla, for there is no fixed base line with which the alinement may be corrected.

4. Lacerations are repaired in the appropriate layers. Catgut mattress sutures are used for the mucosa, catgut for muscle, and medium silk and fine silk for the skin. The skin sutures are usually removed in five or six days.

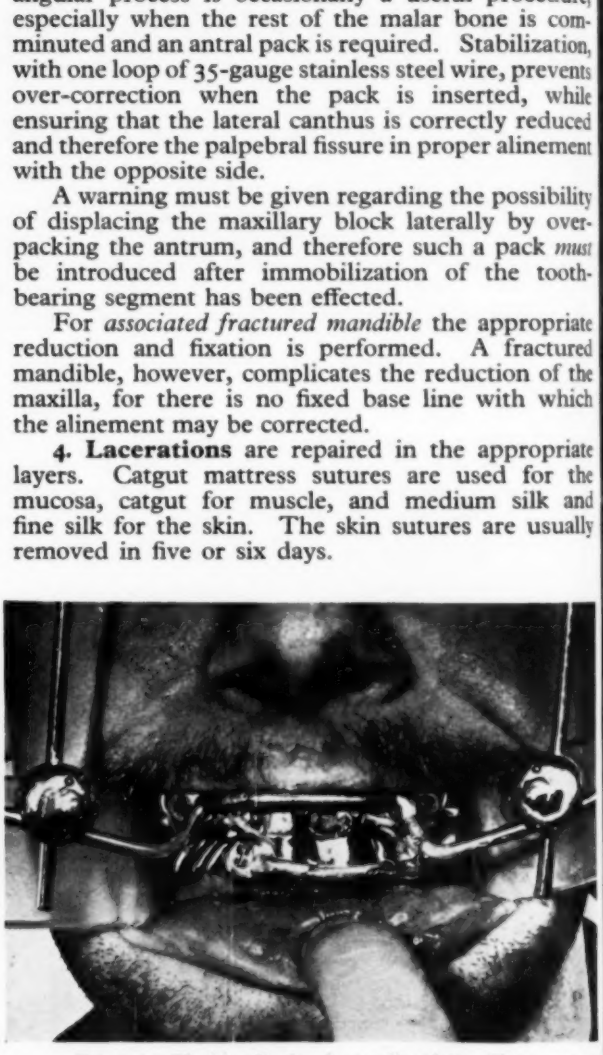


FIG. 322.—Final localization for a split palate. The upper localizing bar has attachments for two vertical frontal bars to the headcap. The lower bar has been used to localize a fractured mandible.

5. The Removal of the Pack and the Application of Intermaxillary Fixation.—The pharyngeal pack is removed, the occlusion finally checked, and intermaxillary fixation applied by means of elastic bands to hooks on the splints. The application of intermaxillary fixation not only ensures correct occlusion, but also maintains absolute immobility

and thus allows resolution of the pterygoid muscle spasm. It must be emphasized that minor adjustments only can be made once the apparatus is in place, as it is a fixation and not a reduction mechanism.

SPECIAL PROBLEMS

1. The Split Palate.—In cases where the teeth are so poor or severely damaged that the patient will inevitably have to wear a denture, a minimal displacement due to a split palate should be accepted. The case is then treated as detailed above, with the proviso that if cheek wire traction is to be used, a palatal bar is added as part of the splint to prevent lateral distraction in the molar regions. The split with displacement is common. It occurred in 28 per cent of cases in this series. It demands separate cap splints which are joined by a bar when reduction has been achieved. In such cases cheek wires are contra-indicated because they exert not only an upward but also a lateral pull which may distract the fragments in the molar regions. A very stout fixation is given by twin frontal rods soldered to the localizing plates and emerging from the angles of the mouth. The disadvantage of localizing the palatal fragments during the operation is that it is time-consuming. To avoid this delay a two-stage fixation may be employed. At the time of operation craniomandibular fixation is used, the palatal elements being reduced to the immobilized mandible and held by intermaxillary fixation. When the patient has recovered from the anæsthetic the fragments are localized by a bar and twin frontal rods used as described above (*Figs.* 310 B, 319, 322).

2. Alveolar Fragments.—These complicated 22 per cent of the cases. If the displacement is minimal and the mandible is intact, and where there is a good complement of teeth, the plaster model of the maxillary teeth may be divided along the fracture line and the fragments positioned in correct relationship to the mandibular teeth. From this corrected model a one-piece splint is constructed and at the

FIG. 323.—On left a patient's denture altered for the three-point maxillary suspension. On right an acrylic base-plate for the endentulous maxilla where a denture was not available.

time of operation the fragments are reduced into it. This technique is of use only if the lesser fragment is freely mobile and should not be employed on any but the slightest displacements.

Where multiple alveolar fragments are present, it may be unwise to attempt to use craniomaxillary fixation as craniomandibular fixation is simpler. The alveolar fragments are reduced to the immobilized mandible and maintained by intermaxillary fixation. These fragments may later be localized, the one to the other, by localizing bars. The commonest alveolar fragment is in the molar region carrying the last two molar teeth. Unless the teeth are subluxated from the fragment extraction should not be attempted, as there is a danger that the whole fragment, which will include part of the floor of the antrum and the pterygoid hamulus, will be removed with the teeth. Large alveolar fragments may be treated as described in cases of split palate.

3. The Edentulous Patient presents a problem in stable fixation, but in such cases it is not of vital

FIG. 324.—Slow forward traction with 4 lb. weight for a case of delayed treatment.

importance from the functional point of view if a slight degree of displacement persists, for this may be compensated for by dentures. If the original upper denture is intact, it may be fitted with loops for cheek wires and a frontal bar attachment (*Fig.* 323). If the dentures are broken an acrylic base-plate with similar attachments is made (*Fig.* 323). The plate is fixed to the maxilla by wires passed through the alveolus, and the operative procedures are carried out as described above.

Craniomaxillary three-point fixation is often the most satisfactory method to use on the edentulous case.

DELAYED TREATMENT

If treatment has been delayed for some two or three weeks the immediate reduction and fixation that has been described is impossible, and slow traction or open reduction is necessary. A weight of 4 lb. is attached to an anterior bar on the upper splint. A guide loop is fixed to the headcap so that the traction, over pulley on a Balkan beam, continues in a constant direction. After three to five days of such traction, the maxilla may be pulled out enough to perform a normal fixation. It must, however, be emphasized that the results of these late procedures are never as satisfactory as treatment within the first few days (*Fig.* 324).

CEREBROSPINAL RHINORRHŒA

Out of 190 cases of middle-third fractures, 45 cases of cerebrospinal rhinorrhœa were encountered (*Table V*). We have had 3 patients referred to us following craniotomies and fascial grafts for cerebrospinal rhinorrhœa consequent upon middle third fractures. Two of these cases continued to show a

leak of cerebrospinal fluid, while the other case developed meningitis. No case of associated fracture of the sphenoidal sinus with rhinorrhœa, as described by Lewin and Cairns (1951), was encountered.

All these patients had identical treatment except for the three aforementioned craniotomies, which

Table V.—A. Incidence and Duration of Cerebrospinal Rhinorrhœa in 190 Cases

Total—45 (24 per cent)	
Average duration for 23 cases	9 days
Longest duration	32 days

B. Complications of Cerebrospinal Rhinorrhœa in this Series

1. Meningitis	2 cases	1 followed fascial grafting—recovered 1 case occurred while awaiting fixation—died
2. Cerebral-aerocele	2 cases	

were performed for associated depressed fractures of the frontal bone. The dural defect with cerebrospinal leak was found on lifting the frontal lobes. In one of these cases the cribriform plate was completely removed. One other case of meningitis occurred, while awaiting maxillary reduction, with subsequent death, while in the third week two of our patients were successfully operated upon by the neurosurgeons for a frontal aerocele.

It is difficult to understand how a detached piece of temporalis muscle or a piece of fascia lata can survive as a graft, and stop the rhinorrhœa, for such a procedure introduces a relatively non-viable substance on to a mobile bed with inadequate support, and indeed, in some cases, with no support at all, and with direct access to infection.

The literature contains numerous references to the treatment of this condition in association with facial fractures. Rowbotham (1949) has recommended delaying treatment if the patient is severely concussed or if the leak stops spontaneously. He also quotes Cone as recommending immediate operation in all compound tears of the basal dura. Calvert (1942) reported 21 cases of cerebrospinal rhinorrhœa in 128 patients, 11 were conservatively treated with 3 cases of meningitis 2 of which died, whilst 1 developed a brain abscess; 10 had a craniotomy and closure of the fistula with 1 fatal case of meningitis. Meredith (1951) advises craniotomy and muscle-graft occlusion if the leak continues for more than ten days.

The question of reduction of the fracture received consideration by Waldron and Balkin (1942), Erich (1942), and Glenn (1938), who recommended waiting seven to fourteen days before reduction to guard against meningitis.

We, however, regard the replacement and immobilization of the fractured bones at the earliest possible moment to be of prime importance to stop the leak, instead of using tissues of extremely doubtful viability to plug the hole (McGregor, 1939).

The floor of the anterior fossa of the skull has been observed during frontal craniotomy, whilst the maxilla of a high Le Fort II type fracture, with associated cerebrospinal rhinorrhœa, is rocked. The cribriform plate and fractured fragments of the neighbouring orbital plates of the frontal bones have been seen to be displaced and overlapping, moving considerably on manipulation of the facial skeleton. Reduction and fixation of the facial fractures will stop the constant movement and shearing of the fragments of the fractured anterior fossa. The edges of a tear in the basal dura may thus come together and heal by scar.

On admission a patient suffering from a leak of cerebrospinal fluid is put on a course of sulphadiazine and systemic penicillin, and this is continued until the rhinorrhœa ceases. The patient is nursed in the sitting position or with the head of the bed on 12-in. blocks. In our practice the majority of fractured faces are reduced and fixed within twenty-four hours of admission, and the average duration of the rhinorrhœa has been ten days from the date of admission. The longest persisting leak lasted thirty-two days. Unless definite signs of meningitis occur no lumbar puncture is performed, and no fluid restriction or dehydrating agents are used. The nose is not irrigated with any antiseptic, and the only local treatment is the removal of accumulated crusts in the nostrils.

POST-OPERATIVE TREATMENT

The patient is usually returned from the theatre with the jaws closed by intermaxillary fixation. A nasopharyngeal tube is therefore necessary until he regains consciousness. Post-nasal bleeding should have stopped and the pharynx should have been inspected for and cleared of blood-clot before the patient is returned to the ward, but a suction apparatus and a soft rubber and gum-elastic catheter and laryngoscope should be at the bedside for the next twelve hours. If respiratory obstruction occurs, which cannot be relieved by suction through the nasopharyngeal tube, the nurses are instructed to cut the intermaxillary elastic bands or wires to get direct access to the pharynx, and to tip the patient's head downward while awaiting the arrival of the medical officer. Although these instructions are given to the nursing staff, it has never been necessary in these 190 patients to divide the intermaxillary fixation. Pethidine, 100 mg., is given intramuscularly as a post-operative drug, but these patients seldom require much sedation once the fractures have been immobilized.

1. Local Treatment

The intra-oral apparatus must be cleaned at least twice daily with 5 per cent sodium percarbonate solution. A Wyatt-Wingrave syringe has been found to be the most suitable apparatus. The occlusion of the teeth must be frequently checked in the early days, the entrance holes of the cheek wires cleaned with 1 per cent cetavlon and dusted with penicillin and sulphathiazole powder, whilst a close watch is kept for any signs of skin necrosis beneath the nasal fixation plates. The tension of the cheek wires must be checked daily, as they may slacken off if the headcap subsides when œdema of the frontal area resolves. Any marked discomfort or looseness of the plaster headcap means that it should be replaced and the apparatus re-assembled.

The intermaxillary fixation can, in many cases, be dispensed with in ten days, but at least three weeks should elapse before the craniomaxillary fixation is loosened and union tested. Slight

'springiness' of the maxillary block may be present at this time and may persist for some days. If no displacement occurs in forty-eight hours the whole fixation is removed.

2. General Treatment

The patient is sat up as soon as he regains consciousness.

In some patients, especially those who develop some post-operative bronchitis or pulmonary collapse, or who have a degree of nasal obstruction, the intermaxillary fixation interferes with satisfactory respiration. A nasopharyngeal tube, lubricated with 2 per cent nupercaine ointment, can safely be left in situ for forty-eight hours, whilst large-bore rubber tubes can be placed in each buccal sulcus. A satisfactory air inflow is usually obtained by using both these methods, and removal of the intermaxillary fixation is seldom necessary to attain a good airway.

The patient is made to do regular breathing exercises. He is sat out of bed on the third post-operative day, and is usually ambulant by the seventh day.

He starts taking a semi-solid diet with purées and fine mince the day after operation, and, when the intermaxillary fixation is released in about ten days, he is given a soft diet. A 3000-calorie diet is aimed at which includes milk, soups, and added Prosol or Casilan, suitably flavoured. Appropriate quantities of vitamins and iron are prescribed.

The unconscious patient with an associated cranial injury is nursed flat, with the head of the bed on 12-in. blocks. Gastric tube feeding must be resorted to in these cases, a total quantity of 1500 c.c. of fluid being given during the twenty-four hours.

POST-OPERATIVE COMPLICATIONS

Post-operative complications were fairly numerous but were generally minor (*Table VI*). Local infection of the coincident mandibular fracture lines, the soft tissues of the face, and the antra, and slight occlusal discrepancies from incomplete reduction were the most frequent. Diplopia, from injury to the orbital floor, epiphora from injury to the nasolacrimal duct, and oro-antral fistulæ following packing of the antrum to control a malar bone were less frequent, while various accidents to the fixation such as cheek wires breaking, temporary alopecia, and pressure sores from the plaster headcaps, and electrolytic ulceration of pin and screw holes or the gingiva were rare.

Table VI.—Post-operative Complications

Local Complications	*Cases*
1. Deranged occlusion	23
2. Infection of the face, antra, and mandibular fractures, etc.	22
3. Diplopia	10
a. Temporary	8
b. Permanent	2
4. Oro-antral fistula	10
5. Epiphora	5
6. Delayed union of mandible	3
7. Accidents to the fixation	6
8. Alopecia (temporary, partial)	3
9. Pressure sores (of scalp)	3
10. Electrolytic ulceration	3
11. Aerocele (cerebral)	2
12. Others	4
General Complications	
1. *Lungs*:	
Pneumonia	2
Collapse of a lobe	2
Foreign body impaction	1
2. *Uræmia*	1
3. *Post-traumatic neurosis* } Associated with fractured skulls	4
4. *Post-traumatic epilepsy* } Associated with fractured skulls	1
5. *Monoplegia* } Associated with fractured skulls	2
6. *Others*	3

There were only 4 cases of post-traumatic neurosis; 1 case of post-traumatic epilepsy; and 2 cases of permanent monoplegia, following upon associated cranial injuries.

Post-operative lung complications were rare, totalling 5 cases only, 1 of which developed bronchiectasis from impaction of a piece of denture in a lower-lobe bronchus.

It is to be regretted that figures showing the degree and permanence of anosmia are not available. It is our impression, however, that the great majority of our middle-third fractures of Le Fort II and III types, with associated fracture and widening of the nasal bridge, have anosmia to a varying degree. A few have a complete and permanent defect, but many have a partial anosmia, which is often variable in a single patient, while not a few state that both smell and taste are present, but are entirely different from their pre-accident sensations.

DEATHS

All 7 deaths (3·7 per cent) occurred within five days of admission, and within nine days of the accident. Three of these deaths were due to gross cerebral trauma, 1 was due to uræmia, 1 to meningitis and bronchopneumonia, 1 to bronchopneumonia and focal necrosis of the liver, and 1 to pulmonary collapse and aspiration of blood. None of these cases had had any operative reduction or fixation.

RESULTS

The assessment of results has been divided into two categories. In relation to *appearance*, 40 per cent were good, comparing well with the pre-accident state, 40 per cent satisfactory with minor change of appearance, while 16 per cent were classified as poor because of bad scars, and flat faces; 4 per cent of the patients died.

In relation to *function*, 80 per cent were satisfactory, 12 per cent had occlusion differing from the pre-accident state by a discrepancy varying from 1 mm. to a complete tooth. Grinding of the cusps in many such cases gave satisfactory function. Three per cent had permanent epiphora in spite of dacryocystorhinostomy, and 1 per cent had permanent diplopia.

CONCLUSION

In conclusion we would like to stress the urgency of the diagnosis of these facial injuries. A good principle is to regard bilateral orbital hæmatomata as indicating a fractured maxilla, and a unilateral hæmatoma as a fractured malar, until these injuries have been excluded by more detailed examination. We have already covered the salient points in the diagnosis.

If the case needs urgent operation for some other injury, an opportunity should be made to put the maxilla into a better position, even although no fixation is available, and an ethmoid reduction will

only take a matter of five minutes at the end of the operation for the associated more serious trauma. Because the patient is unconscious there is no contra-indication to reduction and fixation, indeed it may be an indication to do so, and such procedures should be carried out as soon as possible.

It has already been stated that in severe middle-third injuries the fragments of the floor of the anterior fossa will move when the maxilla is moved. Fixation of the maxilla in a restless and unconscious patient will avoid further trauma to the anterior fossa and the frontal areas of the brain.

To leave a splayed ethmoid block for more than five days is tantamount to accepting its deformity and such an injury should be reduced preferably within twenty-four hours. Early reduction should be performed to control the cerebrospinal rhinorrhœa, while delay will render a maxilla much more difficult to reduce fully without gradual traction-reduction. If delayed longer, then open operation and osteotomy may have to be performed.

It is urged that special centres, equipped both surgically and dentally to deal with these injuries, be *allowed to have these patients as early as possible* after their accidents. In this way maximum function and good appearance can be restored to them (*Table VII*).

Table VII.—A. AVERAGE TIME INTERVAL BETWEEN THE ACCIDENT AND DEFINITIVE TREATMENT

1940–45	11 days
1946–51	4 days

B. AVERAGE STAY IN THE PLASTIC AND JAW UNIT

1940–45	80 days
1946–51	37 days

Table VII correlates the time between the accident and definitive treatment with the duration of hospitalization.

SUMMARY

One hundred and ninety cases of fracture of the middle third of the face admitted to a single unit have been analysed. The recommended classification is that of Le Fort. The diagnosis and treatment of the various fractures are described, and a conservative attitude to the problem of cerebrospinal rhinorrhœa in these injuries is advised.

We wish to record our gratitude to Mr. Rainsford Mowlem, Director of the Plastic and Jaw Unit, Hill End Hospital, St. Albans, for his great help; to Mr. B. W. Fickling, Dr. A. B. Macgregor, and our other colleagues of the same unit, under whom many of the cases analysed were admitted; and to Miss Walker for the photography.

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OBLITERATIVE ARTERIAL DISEASE IN A YOUNG FEMALE

A POSSIBLE CASE OF THROMBO-ANGIITIS OBLITERANS ASSOCIATED WITH DIGITAL GANGRENE AT BIRTH

BY DAVID MESSENT

FROM THE DEPARTMENT OF SURGERY, POSTGRADUATE MEDICAL SCHOOL OF LONDON

NEONATAL gangrene and occlusive arterial disease in a young female are, in themselves, both conditions of sufficient rarity to be of considerable interest. The occurrence of both in the same patient makes a fascinating clinico-pathological problem.

CASE REPORT

A young housewife, aged 36, attended the out-patient department of Hammersmith Hospital with symptoms of peripheral vascular disease. Her story started at birth when, as far as can be ascertained, four toes were black and had to be amputated at the age of 2 weeks. She was unaware of any other abnormality as regards her birth. The distal half of the foot was always cold and blue, but she was not worried unduly by this until the age of 32, when she commenced to have pain in the left foot. This pain came on after walking about a hundred yards, was relieved by rest, but returned almost immediately on the resumption of walking.

This symptom persisted and in 1949 the remaining toe on that foot became ulcerated. In spite of conservative treatment this failed to heal and in June, 1950, she was admitted to hospital. At that time she had wasting of the left calf, her left foot was cold and cyanosed, and the remaining toe was ulcerated.

Nutrition of the foot was fair and the posterior tibial and dorsalis pedis pulse were palpable in both feet. The right foot was in all aspects normal. Whilst awaiting admission to hospital she was treated with 'Priscol' (50 mg. t.d.s.) with a good response.

On June 29, 1950, a left lumbar sympathectomy was performed and the fifth toe was amputated. Her recovery was uneventful, the foot healing by first intention; the symptoms in her left foot were relieved entirely and have never recurred, the lower extremity remaining warm and dry.

In February, 1951, she developed similar symptoms in her right foot—pain after walking a hundred yards which was relieved by rest, coldness, whiteness, and

numbness. No ulceration or gangrene occurred, and at no time did thrombophlebitis occur, nor had she claudication in her calf muscles. Apart from an attack of whiteness there had been at that time no symptoms in the hands.

Family History.—Her father, at the age of 40, developed gangrene of a little toe, which, after prolonged treatment, ended with a below-knee amputation. He lived then until 72, when he died of "heart disease". No other member of her family gives a history of heart disease.

Personal History.—She smokes ten cigarettes a day.

ON EXAMINATION.—She was a healthy-looking young woman who showed no abnormality except in the extremities. Her blood-pressure was 110/70. The hands presented no evidence of wasting or skin atrophy. Both radial arteries were easily palpable, but neither ulnar artery could be felt. Allen's test (Allen, 1929) was positive. The left calf was wasted and the left foot, from which all the toes were absent, was warm and dry. The right foot was cold, there was little evidence of skin atrophy; on dependency there was slight rubor and, on elevation, pallor. The peripheral arterial pulses were :—

	Right Leg	*Left Leg*
Femoral	+	+
Popliteal	+	+
Posterior tibial	0	0
Dorsalis pedis	0	0
Oscillometric deflections were :—		
Above the knee	5½	4
Mid-calf	4	3
Ankle	1	2

INVESTIGATIONS.—

Blood: *Hb*. 13·5 g. (91 per cent).
W.B.C. 10,000 per c.mm.
Differential Count

	Per cent	*Per c.mm.*
Polymorphonuclears	72	7200
Eosinophils	2	200
Basophils	1	100
Lymphocytes	15	1500
Monocytes	10	1000

Cold Agglutinin Titre
Titre 2–5° C. 8
" 20° C. } Negative
" 37° C. }
No evidence of cryoglobulinæmia.
W.R. Negative.

Urine: No sugar. No albumin.
No hæmoglobinuria.
17 ketosteroids: Total for 24 hours: 6·0 mg.
Creatinine: Total for 24 hours: 720 mg.

E.C.G.: Within normal limits.

Radiographs: *Chest*: Calcareous nodules in right root but no recent infiltration. Cardiac outline not abnormal.
Lower limbs: No arterial calcification.
Left foot: Absence of toes form metatarsal joints. Some disuse decalcification.

Arteriography (*Fig*. 325): Percutaneous (50 per cent Pyelosil).

Left leg: The superficial and deep femoral arteries are of normal calibre and smooth in outline, but there are a few tortuous collaterals in the lower thigh which must be considered abnormal. The popliteal artery appears normal. The anterior tibial artery fills well throughout its length and shows no evidence of atheroma, but the dorsalis pedis artery does not fill. The posterior tibial is seen well in its upper half and appears normal; its lower half, however, is probably diseased. The plantar artery is filled largely by collateral vessels. There are many fine vessels apparent in the lower third of the leg whose presence is abnormal. The veins are well demonstrated in the leg.

Right leg: The superficial and deep femoral arteries appear normal but here also there are a few tortuous collaterals in the lower thigh. The popliteal artery shows no atheroma. The upper part of the anterior tibial artery fills normally but the lower half is suspect. The posterior tibial artery fills well in its upper third but the distal two-thirds are represented by a line of fine tortuous vessels. There are many fine collaterals in the lower half of the leg. The dorsalis pedis is patent only for the proximal half of its length, but the plantar artery shows no block. There is no venous filling.

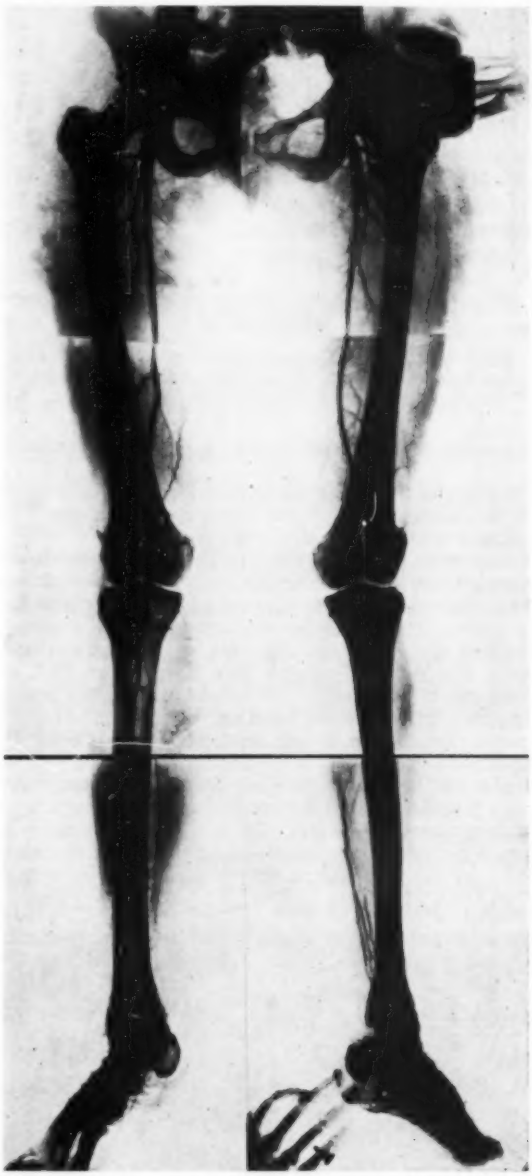

FIG. 325.—Bilateral femoral arteriogram. The foot has been externally rotated for the third film of each leg and accurate continuity in the composite picture is not possible. A black line has therefore been inserted between the second and third film in the reproduction.

A reflex heating test (*Fig*. 326) was carried out but after heating the body for sixty minutes the increase in

temperature in the right great toe was only 1·75° C. In view of a room temperature of only 17° C. this could not be accepted as proof of inability to obtain vasodilatation because of organic arterial obstruction. No alteration

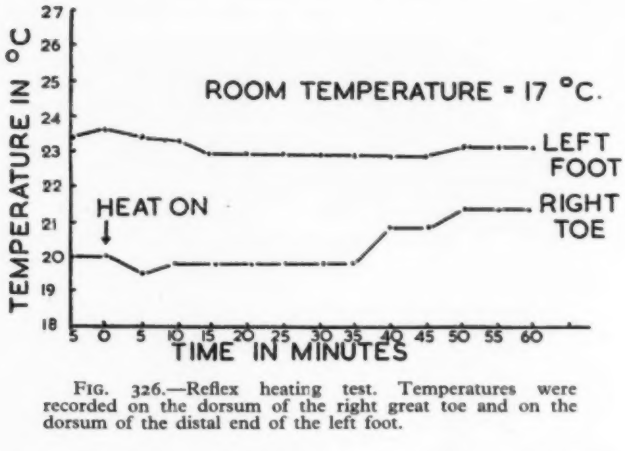

FIG. 326.—Reflex heating test. Temperatures were recorded on the dorsum of the right great toe and on the dorsum of the distal end of the left foot.

of temperature occurred in the sympathectomized left foot.

An exercise test using the venous occlusion plethysmograph to measure the calf flow was also carried out. This was almost within normal limits (*Figs.* 327).

TREATMENT.—On Sept. 20, 1951, a right lumbar sympathectomy (Mr. Peter Martin) was performed and at the same time a biopsy was taken of the right dorsalis pedis artery, which was known to be non-functioning.

Following operation she made an uninterrupted recovery and was discharged from hospital on Oct. 1.

FOLLOW-UP (Oct. 24).—The pain in the right forefoot has improved but has not been relieved completely. She is complaining of pain in her right calf which is suggestive of claudication. This is a new symptom. For one week she has been experiencing Raynaud's phenomenon in three fingers of her left hand.

HISTOLOGICAL REPORT (Dr. C. V. Harrison).—The specimen included three pieces of artery, all approximately 0·5 mm. external diameter, two veins of similar size and

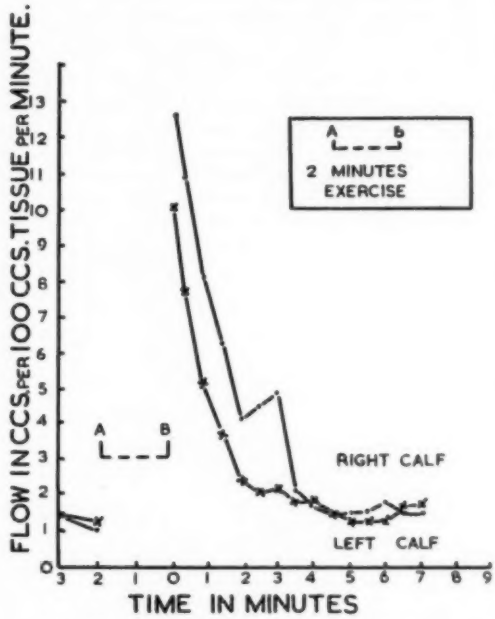

FIG. 327.—Exercise tolerance test. The calf flow in response to exercise has been measured with the venous occlusion plethysmograph. Between A and B arterial occlusion was applied and the calf muscles put through graduated exercises. At B the arterial occlusion was released, exercise stopped, and measurements of the calf muscle flow made. This response is slightly less than would be expected in the normal.

and is thrown into deep folds (*Fig.* 328). The media is intact and is contracted, appearing thickened. It is traversed by numerous enlarged vasa vasorum (*Fig.* 329). The adventitia is thickened by fibrosis and contains an excessive number of vasa vasorum (*Fig.* 329). There is a similar fibrous thickening surrounding the nerve. The two veins are unaffected.

This is the scarred stage of an earlier active lesion and it is, therefore, not possible to make a dogmatic

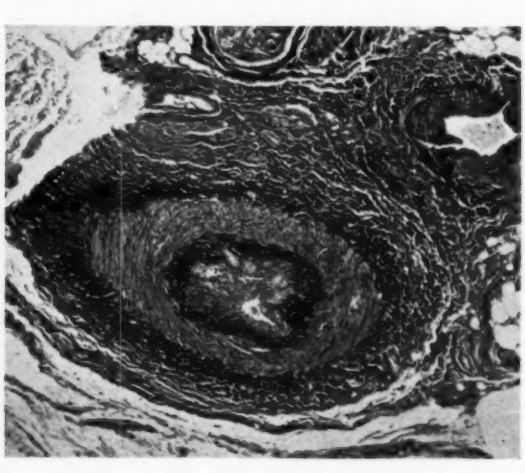

FIG. 328.—Biopsy of dorsalis pedis artery and associated vein. (× 45.)

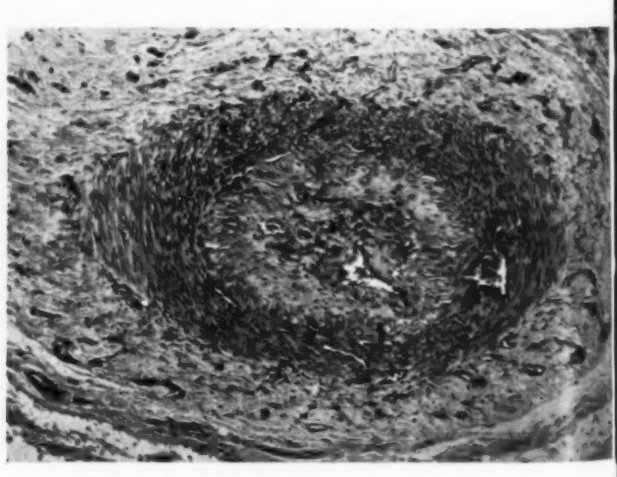

FIG. 329.—Biopsy of dorsalis pedis artery. (× 67·5.)

one nerve. All three arteries show the same change and may be three pieces of one vessel. The lumen is occluded by fibrous connective tissue containing a few well-formed arterioles and venules and a few lymphocytes and histiocytes (*Fig.* 329). The internal elastic lamina is unbroken

diagnosis. There is nothing to suggest atherosclerosis; giant-cell arteritis and polyarteritis nodosa are unlikely in the presence of an intact media and internal elastic lamina. In my opinion this is the healed stage of thromboangiitis obliterans (Buerger's disease).

DISCUSSION

Gangrene in infancy and childhood due to arterial insufficiency is rare but has been described in the literature on several occasions. Gross (1945), quoting Martini as having described a case in 1828, summarizes 47 cases from the literature in children varying in age from newborn to 13 months old. By far the majority of these occurred within the first twenty-one days of life, and 4 of them on the first day. In discussing aetiology he suggests five possibilities: (1) sepsis, (2) trauma, (3) emboli from congenital heart disease, or from (4) the thrombosing ductus arteriosus, or from (5) the obliterating umbilical arteries: and to these might be added (6) exposure to cold at birth, (7) local thrombosis of the vessels in the digits. Aird (1949) records 3 cases of bilateral gangrene of obscure origin occurring in infancy and Lewis (1949) recognized bilateral gangrene of the digits occurring in children commonly associated with malnutrition or ill health. The exciting factor of the intimal damage which gave rise to thrombosis and gangrene was not apparent.

In the present case sepsis, trauma, local thrombosis, and exposure to cold as cause of the neonatal gangrene can, at this stage, no longer be confirmed or excluded. Gross has adduced clear evidence that some form of embolism was the likeliest cause of the neonatal gangrene in some at least of his cases. The neonatal incident in the present case resembles Gross's first case, in which he suggests that the loss of three toes was due to emboli lodging in the tarsal and arcuate branches of the dorsalis pedis and in the lateral plantar artery. In his case the dorsalis pedis pulsation could be felt on the proximal part of the foot, as it was in this patient when first seen at the age of 36. However, it is perhaps apposite to mention in this connexion that in at least one of Aird's cases of neonatal gangrene, digital pulsation was palpable in an affected digit during the presence of gangrene. Gross has also shown that when iliac or femoral embolism has occurred in an infant with loss of all pulses in the limb, these pulses may later return. But in the present case arteriography in the adult shows a patent femoropopliteal trunk so normal that it can hardly have been the seat of embolism and later canalization.

Whatever the cause of the neonatal gangrene in this case, its relation to the occlusive disease which she has developed in adult life requires close consideration. One fact is beyond dispute: the patient was left with a cold, blue foot and therefore presumably with some residual vascular insufficiency of the left foot from birth. Three alternatives have to be considered: (*a*) the present condition is a sequel of the neonatal incident; (*b*) the infantile gangrene of the toes was in fact the first manifestation of the adult disease; (*c*) two entirely different and separate pathological processes have occurred in the same patient.

Each of these must be considered in the light of the evidence available and the recognized modern pathological and clinical classifications of arterial disease.

a. As a Sequel of the Neonatal Incident.—As a result of embolism, thrombosis, or other occlusive effects at birth, can a thrombosis have spread later to affect a proximal part of the arterial tree? In the first place it is unlikely that such an extension would take place after an interval of thirty-two years, and even if such an explanation were acceptable in respect of the left leg, in the case of the right leg it is untenable as this limb was symptomless until she was 36 years old.

b. The Neonatal Incident was the Beginning of the Adult Condition.—If this is to be accepted, then it must be assumed that at birth there was present one of two conditions: Raynaud's disease (with early tissue loss) or thrombo-angiitis obliterans.

Raynaud's disease as the sole cause of the neonatal and adult gangrene: It is well known that tissue loss can occur with varying rapidity in an adult with Raynaud's disease, but to assume that a newborn child could develop gangrene due to Raynaud's disease would be startling. Nevertheless, this patient has manifested Raynaud's phenomenon in the left foot for as long as she can remember, with the added incident of the loss of her remaining toe from gangrene at the age of 36. Such an explanation could conceivably be accepted for the right leg. The clinical absence, one year later, of distal pulses in both legs, however, no less than the arteriographic demonstration of obliterative disease of the calf vessels, is quite inconsistent with a diagnosis of Raynaud's disease.

Thrombo-angiitis obliterans as sole cause of the neonatal and adult gangrene: The presence of thrombo-angiitis obliterans on the day of birth would seem to be unprecedented and a history that the patient's father had a leg amputated at the age of 40 for gangrene of a toe, whilst interesting in the highest degree, cannot be advanced as evidence of inheritance.

c. Two Entirely Different and Separate Pathological Processes.—Neonatal gangrene and obliterative arterial disease in a young adult woman are both of such rarity that for both to occur in the same patient without mutual relationship would seem to be an extravagance of hypotheses. Nevertheless this eventuality must be considered; that neonatal gangrene of some unknown and unrelated cause was followed in later life by Raynaud's disease or thrombo-angiitis obliterans. I have already reasoned on clinical and arteriographic grounds against Raynaud's disease as a cause of the adult symptoms, but thrombo-angiitis has not yet been excluded as a cause of the adult phenomena.

Thrombo-angiitis obliterans (Buerger's disease or juvenile obliterative arteritis—Boyd, Ratcliffe, Jepson, and James, 1949) is an episodic disease commencing typically in young male adults with arterial obliteration beginning peripherally and associated in 40 per cent of cases with thrombophlebitis migrans (Allen, Barker, and Hines, 1946). This patient is a woman. Buerger's disease in women is extremely rare, as may be seen from *Table I*. The overall incidence in women in 3620 cases was 0·9 per cent, though it is suggested by Silbert (1948) that the incidence is on the increase. These are large American and Japanese series based on a clinical diagnosis only, and it is not possible to find a comparable series from this country. There is no doubt, however, that the diagnosis of thrombo-angiitis obliterans is made in Britain with considerably greater caution, and, even in the male sex, it is a comparatively rare

disease. In 1951 Fisher, Zukerman, and Sweeney cited from the literature only 7 cases of thrombo-angiitis obliterans in the female which had been proved histologically, and added 1 case of their own.

In the present case there is strong presumptive histological evidence of this condition. Clinically,

Table I.—INCIDENCE OF THROMBO-ANGIITIS OBLITERANS IN WOMEN. CLINICAL DIAGNOSIS

NAME OF AUTHOR	DATE OF PUBLICATION	NO. OF CLINICAL CASES OF THROMBO-ANGIITIS OBLITERANS		
		Total No. of Patients	No. of Female Patients	Female Patients as Percentage of Total
Koyono	1921	120	11	0·8
Buerger	1924	500	2	0·4
Horton and Brown	1932	700	10	1·4
Herrel and Allen	1936	350	1	0·2
Silbert	1935	1200	2	0·1
Williams and Barker	1946	750	2	0·2
Silbert	1948	600	23	4·0
Total		4200	41	0·9

the course of the disease has been marked by episodes at birth: at the age of 32 when pain started in the left foot; the loss of her last left toe at the age of 36; the onset of pain and Raynaud's phenomenon in the right foot and the disappearance of all peripheral pulses in both feet; finally the suggestion of involvement of her hands. There has been no thrombophlebitis migrans.

All these features favour a diagnosis of thrombo-angiitis obliterans, but there is the inescapable fact—she had neonatal gangrene followed by Raynaud's phenomenon which persisted throughout childhood and adolescence until her occlusive disease developed in early adult life.

CONCLUSION

Here, then, is a young woman with neonatal gangrene and early adult obliterative arterial disease, both of such great rarity that their inter-relationship here can hardly be denied. The evidence available depends so largely on the history obtained rather than observation that it is probably better to describe this patient's condition as one of unilateral neonatal gangrene followed by attacks of the Raynaud's phenomenon, and in early adult life by bilateral occlusive arterial disease, rather than to try to fit it into any known classification of arterial disease in present use.

SUMMARY

1. Unilateral gangrene at birth, Raynaud's phenomena throughout life, and the development of bilateral obliterative arterial disease in early adult life, are recorded here as occurring in the same female patient.

2. The aetiology of the neonatal gangrene and its relationship to the Raynaud's phenomena and occlusive arterial disease is discussed. The diagnosis of the adult condition is considered.

3. Whilst the clinical and histological findings in the adult condition favour a diagnosis of thrombo-angiitis obliterans, the neonatal gangrene and Raynaud's phenomena can hardly be discounted as unrelated. It is concluded that it is better not to try to fit the case into any known classification of arterial disease in present use.

I am grateful to Dr. C. V. Harrison for his report on the biopsy. The microphotographs and arteriographic reproductions are the work of Mr. E. V. Wilmott, F.I.B.P., F.R.P.S., and Mr. R. J. Marshall respectively.

Addendum.—In the interval between writing and publication, the obliterative process has advanced proximally. The popliteal pulse is now absent on on either side and the patient is suffering from intermittent claudication in both calves. There has been no further evidence of disease of the hands.

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DIFFUSE SYSTEMIC ANGIOMATA

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It is generally accepted that angiomata are the commonest of all tumours, if indeed they can be called tumours at all. A cursory examination of the *Index Medicus* will reveal that dozens of papers are written annually on this subject, but despite this spate of literature there remains a group of angiomata which cause difficulty in diagnosis and often pass unrecognized. These are the deep angiomata involving muscle. The first of these tumours was described by Liston in 1843 involving the semimembranosus muscle, and since that time fresh cases have been reported and excellent reviews of the literature have been made by Davis (1908), Davis and Kitlowski (1930), Jenkins and Delaney (1932), Elkin and Cooper (1947), and many others. These authors have all referred to hæmangiomata in striated muscle, but it has been found that the abnormality affects surrounding tissue also, including bone and subcutaneous tissue, and there is little justification for separating the muscular tumours from others in the same group. Usually the skin escapes, and it is for this reason that the diagnosis is often missed. Watson and McCarthy (1940), in a careful analysis of more than 1000 cases, produced the following classification of the lesions :—

Classification :—

Capillary—usually superficial and well circumscribed.
Angiolipoma.
Angiofibroma.
Spider nævus.
Cavernous—larger and more extensive and quite commonly mixed with capillary type ;
Angioblastoma—hyperplasia of lining cells with production of a semi-solid tumour ;
Racemose (cirsoid) angioma : pulsating ;
Diffuse systemic angioma ;
Metastasizing angioma ;
Port-wine stain ;
Hereditary hæmorrhagic telangiectasis.

The diffuse systemic angioma is the one which is least well recognized. It is a tumour arising from malformation and proliferation of pre-existing vessels in muscles, glands, bone, and viscera rather than a true neoplasm. In some areas of the same tumour there will be the formation of capillaries, in other areas cavernous systems, while in others the formation of large venous spaces. These tumours rarely communicate freely with the general systemic circulation and do not commonly show the signs of increased blood-flow, warmth, pulsation, or bruit which are seen in the racemose or cirsoid angioma. These may well be called by Albrecht's name of hamartoma, a disturbance or disorderly type of growth of otherwise normal tissue occurring in a normal place.

The purpose of this paper is to report 16 cases of diffuse systemic angiomata, some involving the whole limb and others involving large groups of muscles, or glands, which illustrate the way in which a so-called non-malignant tumour extends from one muscle group to another without having definite boundaries, and to consider methods which may be adopted for their treatment.

CASE REPORTS

Case 1.—C. B. L., female, aged 5 months. *Hæmangioma of masseter and facial muscles.*

The child was brought to hospital with a mass over the left side of the face, 2 in. in diameter, ill-defined, fluctuant, not attached to skin but not freely movable over underlying structures. A small capillary angioma was present in the skin over the tragus. X-ray examination showed no bony change in the mandible and no phleboliths.

Treatment.—Injection with neovaricaine at multiple points in the tumour.

Result.—Satisfactory after an acute inflammatory reaction for several days.

Case 2.—C. B., female, aged 6 years. *Hæmangioma of infraspinatus.*

Complained of a slowly growing mass over left scapular region since birth. Not interfering with movement. The mass was 4 in. in diameter, lobulated and fluctuant, not attached to skin, but not moving freely on deep structures.

Pre-operative Diagnosis.—Lipoma.

Treatment.—Exploration. A large hæmangioma was found arising from infraspinatus and was excised.

Result.—Satisfactory.

Histology.—A large amount of fibrous tissue mixed with blood-vessels and blood-spaces with considerable cellular poliferation in the walls of the spaces.

Case 3.—F. M. F., female, aged 23 years. *Hæmangioma of sternomastoid.*

Complained of a slowly growing mass in the neck since birth. There was a rounded cystic mass 2 in. by 1½ in. in front of the upper border of sternomastoid, soft, fluctuant, but not translucent. Not attached to skin, but not moving freely on deep structures.

Pre-operative Diagnosis.—Branchial cyst.

Treatment.—Excision. A rounded cavernous hæmangioma was found arising from the anterior part of sternomastoid and involving platysma. The whole mass was excised.

Histology.—The mass consisted of large and small blood-spaces interspersed with loose connective tissue.

Case 4.—W. F., male, aged 14 years. *Hæmangioma of whole lower limb.*

Complained of multiple cystic swellings over the right leg since birth. There was some discomfort during prolonged walking. On examination, there were multiple soft cystic swellings over the foot, knee, and groin (*Fig.* 330 A). The swellings could be emptied by pressure and refilled spontaneously. Fluid could be pressed from one swelling to another, but it was not possible to evacuate them all simultaneously. An Esmarch bandage wound up from the foot caused the swelling at the knee to increase, and as it was taken higher the swelling at the knee was compressed and the one in the groin enlarged and became quite tense. There was no pulsation. X-ray showed phleboliths in the thigh and involvement of patella, medial condyle of femur, tibia, and fibula, os calcis, astragalus, and first metatarsal by hæmangioma (*Fig.* 330 B).

TREATMENT.—No treatment short of amputation could get rid of such an extensive angioma, and as function was not seriously impaired, this was not indicated.

A B

FIG. 330.—*Case* 4. A, Hæmangioma of whole lower limb. B, Phleboliths in the thigh with extensive bony involvement.

Case 5.—T. L. H., female, aged 15 years. *Hæmangioma of trapezius.*

Complained of a small hard mass on the back for twelve months. A mass 1 in. in diameter was situated between the spinous process and the medial border of the right scapula. Not attached to skin, but not freely movable on deep structures.

PRE-OPERATIVE DIAGNOSIS.—? Neurofibroma.

TREATMENT.—Excision. A mass was found involving trapezius. Fairly well encapsulated.

HISTOLOGY.—Many poorly formed blood-vessels with walls of varying thickness on a fibrous-tissue stroma. One larger space filled with blood-clot.

Case 6.—S. C. Y., female, aged 18 years. *Hæmangioma of small muscles of hand.*

Complained of repeated attacks of pain in the right hand for six months. She was aware of bluish discoloration and swelling of the hand which had been present at birth and had formerly become less when the hand was raised. There had been occasional pain for a very long time, but not so severe as in the last six months. On examination, the dorsum of the hand, all fingers, and the centre of the palm were covered by bluish soft swellings with irregular areas of thickening in them. All finger movements were free, but the swellings became more tense when the hand was clenched. The swellings were partially reduced in size by elevation of the hand. The skin showed no angiomatous change. Radiography showed considerable bony involvement (*Fig.* 331).

TREATMENT.—Diathermy coagulation using an insulated needle was attempted on three occasions, but was only partially successful. The whole lesion was therefore treated with interstitial irradiation with fair success.

Case 7.—L. C. M., female, aged 13 years. *Hæmangioma of vastus intermedius.*

Complained of a tender lump in the medial aspect of the thigh for one year. Three years ago she had an injury to the thigh followed by abscess formation, and since then the thigh was slightly swollen and painful. There had only been a definite lump, however, for one year. On examination, there was a firm oval lump 1½ in. in diameter over vastus intermedius, not attached to skin but not freely movable, and becoming fixed as the extensive quadriceps was contracted.

A B

FIG. 331.—*Case* 6. A, Hæmangioma of fingers and small muscles of hand. B, Extensive involvement of proximal phalanges of the index finger.

PRE-OPERATIVE DIAGNOSIS.—? Neurofibroma. ? Lipoma.

TREATMENT.—Excision. The tumour was found to originate in vastus intermedius and was completely excised. Histologically it consisted of large and small blood-spaces interspersed with loose connective tissue.

Case 8.—L. C. S., male, aged 19 years. *Hæmangioma of subcutaneous tissue of forearm.*

Complained of a lump over the left forearm for ten years, painful during the last month. The mass had grown progressively larger. On examination, a softish swelling was present on the lateral aspect of the left forearm just above the wrist. The swelling was fluctuant and was not attached to skin nor firmly attached to deep structures. Wrist movements were normal. X-ray examination showed no bony lesion in radius or ulna. Aspiration produced pure blood.

TREATMENT.—Excision. The mass was subcutaneous and consisted of many dilated vessels, some filled with clot. It was completely excised.

Case 9.—C. M. S., male, aged 12 years. *Hæmangioma of biceps and brachialis.*

Complained of a swelling over the left arm for three years. A swelling had actually been present since birth, but had only been troublesome and painful during the last three years, when it increased in size. On examination, an oval swelling 2 in. by 1½ in. was present on the front of the left arm below the main part of the belly of the biceps. It was soft, fluctuant, with an ill-defined edge, and dilated veins could be seen over the surface. There was no pulsation in the tumour and no bruit. X-ray examination normal. Venogram demonstrated no connexion between the superficial veins and the tumour. Aspiration produced pure blood.

TREATMENT.—Excision. With a tourniquet in place the mass was explored and found to be an extensive hæmangioma involving both biceps and brachialis. A good deal of clot was present in the vessels. The mass was excised as completely as possible and bleeding controlled by a gelfoam pack and firm bandaging. The result was satisfactory, although flexion of the elbow was not very strong. Section showed that the mass consisted of well-formed vessels of all sizes, some filled with blood-clot.

Case 10.—T. Y., female, aged 10 years. *Hæmangioma of gastrocnemius.*

Complained of a painless swelling of the calf for three years, steadily increasing in size. No history of trauma. On examination, the left calf was bigger than the right. The swelling was diffuse without definite boundaries, not lobulated, not fluctuant, not attached to skin. X-ray examination showed no abnormality in tibia or fibula.

PRE-OPERATIVE DIAGNOSIS.—Intermuscular lipoma.

TREATMENT.—The leg was rendered avascular with an Esmarch bandage and tourniquet. An incision was made over the mass. The gastrocnemius and soleus were partially replaced by a pink structureless mass. A piece was taken for section. Histologically the tumour consisted of masses of newly formed capillaries in loose fibrous tissue and resembled a capillary angioma.

Case 11.—K. T. K., female, aged 48 years. *Hæmangioma of latissimus dorsi and erector spinæ.*

Complained of a rapidly growing mass in the right flank for three years. Three years ago patient was admitted to hospital, when splenectomy was carried out for polycystic spleen. Recovery was uneventful. At that time there was a small capillary angioma involving the skin of the right flank. Since then it had grown rapidly and produced a mass 9 in. in diameter and 4 in. thick. The mass was nodular, firm in places and soft in others. The skin over the centre was discoloured and attached to the mass. The mass was also attached deeply. X-ray examination showed multiple phleboliths in the mass.

TREATMENT.—Excision. The mass was partially excised. It was found to involve latissimus dorsi and the erector spinæ group of muscles. The mass contained a lot of solid tissue interspersed with blood spaces. Histologically the mass consisted of fibrous and fatty tissue containing many new blood-vessels.

Case 12.—S. S. K., female, aged 3 years. *Hæmangioma of fingers.*

Complaining of swelling of the index and middle fingers of the left hand since birth. There was no pain

FIG. 332.—*Case* 14. Soft-tissue swellings in suprasternal notch and over right parotid gland.

and no recent enlargement. On examination the middle finger was grossly distended by a subcutaneous hæmangioma and the underlying phalanges were irregular in shape. A small hard loose mass could be felt in the tissue. Movement was not interfered with. The index finger was similarly affected but to a lesser degree. The skin of both fingers was not involved. X-ray examination showed involvement of phalanges of both fingers, index worse than middle, and two small phleboliths.

TREATMENT.—Interstitial irradiation with radium needles.

Case 13.—L. L., female, aged 24 years. *Hæmangioma of tibialis posterior.*

Complained of a swelling in the lower part of the calf for several years, slowly increasing in size, and painful on exercise. There was no history of trauma. On examination, a diffuse swelling was seen in the lower calf deep to the tendo Achillis. It was soft but not fluctuant. It was less readily felt when the muscle was tightened. X-ray examination showed no abnormality. Aspiration produced dark blood.

PRE-OPERATIVE DIAGNOSIS.—Hæmangioma.

TREATMENT.—The mass was explored and found to be a hæmangioma involving the tibialis posterior and the neurovascular bundle. It was partially excised. Histology showed fibrous tissue with many new blood-vessels.

Case 14.—C. C. Y., male, aged 32 years. *Hæmangioma of platysma, masseter, and parotid gland.*

Complained of slowly growing masses in the preauricular and suprasternal regions for more than twenty years. There was no pain, no history of trauma, and no

other complaints. On examination, a soft swelling 2 in. in diameter was present in the pre-auricular region. The borders were regular, the skin normal but slightly bluish, and many small nodules were felt on deep palpation. A similar swelling was present above the suprasternal notch (*Fig.* 332). X-ray examination showed multiple phleboliths.

PRE-OPERATIVE DIAGNOSIS.—Hæmangiomata.

TREATMENT.—The lower mass was excised completely. The upper mass was explored and found to

FIG. 333.—*Case* 15. Hæmangioma of deltoid with multiple phleboliths.

involve the parotid gland and masseter. It was partially excised. A parotid fistula resulted and was cured by irradiation.

Case 15.—C. H., male, aged 26 years. *Hæmangioma of deltoid.*

Complained of a swelling over the right shoulder for twenty years, enlarging slowly and often painful, especially on moving the shoulder. It was first noticed in childhood after being hit with a stick. On examination, a soft bluish swelling covered by normal skin was present over the front of the right shoulder. Many small hard nodules were palpable. X-ray examination: many phleboliths seen (*Fig.* 333).

PRE-OPERATIVE DIAGNOSIS.—Hæmangioma.

TREATMENT.—Excision. Many blood spaces were filled with clot. The mass was completely excised.

Recovery was satisfactory and movement of the shoulder free and painless.

Case 16.—Y. P. M., male, aged 19 years. *Hæmangioma of kidney and psoas.*

Complained of intermittent hæmaturia for five months. Bleeding was profuse and clots were present. The attacks lasted about one day and occurred at intervals of one month. On examination, physical examination was completely negative. The urine contained R.B.C.s, but was otherwise normal. X-ray examination of kidney showed no abnormality in direct film or intravenous pyelography. Cystoscopy: Bladder normal. Bright blood was seen emerging from the left ureter. Catheters were passed up both ureters without difficulty and retrograde pyelography was normal.

DIAGNOSIS.—? Hæmangioma of kidney.

TREATMENT.—Exploration of kidney. The kidney was normal in size and appearance. A large hæmangioma was found involving the left psoas muscle. The ureter was not, however, affected. Nephrectomy was performed, but the angioma in psoas left alone. The specimen showed several angiomata involving the pelvis of the kidney.

Recovery was uneventful and there was no further bleeding.

DIAGNOSIS

(*Table I*)

Consideration of the foregoing case histories brings out certain points which help in diagnosis. Most of these patients are young. In this short series only 2 were over 30 and Oughterson and Tennant (1939), discussing hæmangioma of muscles, say that 95 per cent occur below the age of 30. The symptoms complained of are swelling and pain. The swelling is commonly present over a long period of time, but for some reason the mass starts enlarging more rapidly. In some instances this is the result of trauma, and although this was not a prominent feature in the cases reported here, Jenkins and Delaney (1932) found that it is related to enlargement of the tumour in 17 per cent of more than 200 cases reviewed. Pain is a fairly common symptom and usually occurs in cases in which there has been thrombosis.

Skin involvement is relatively uncommon in the diffuse angiomata and was seen in only 2 of the 16 cases reviewed.

Functional impairment is common when muscle involvement is extensive and there is limitation of movement, muscle weakness, and, when the leg is involved, a limping gait.

Radiological evidence is sometimes of value in making the diagnosis. A soft film will often show a soft-tissue shadow resembling tortuous dilated vessels and phleboliths may be seen. A picture of higher penetration will show bone involvement on occasions. Venography may be of value in making a diagnosis, but the tumour will not always fill as the communications with the general circulation are scanty and the blood in the tumour is stagnant. The most conclusive evidence, however, is obtained by aspiration of the tumour when pure blood will be obtained.

Despite all these considerations the diagnosis may be quite difficult to make if the tumour involves muscle only, and in the cases reviewed by Jenkins and Delaney the diagnosis was correctly made in only 21 out of 256 patients. In the present series we were more fortunate because of frequent subcutaneous involvement. The differential diagnosis is usually between a hæmangioma and an intermuscular lipoma.

TREATMENT

Hæmangiomata have been treated in many different ways and no one way is suitable for all cases. The following are the common methods in use at the present time:—

1. Carbon-dioxide snow.

2. Injection of coagulants.
3. Irradiation: Radium—surface, interstitial; X rays.
4. Ligation of vessels.
5. Excision.

Carbon-dioxide snow is very suitable for lesions involving skin only, but is quite unsuitable for the diffuse systemic angiomata considered here. The injection of coagulating fluids is not entirely satisfactory. It produces thrombosis and a moderate inflammatory reaction, and if the lesion is extensive the treatment must be done in stages. In the 16 cases reported, this method was used only once, but on several occasions it has been used in more superficial angiomata without great success. Irradiation with interstitial radium needles has in this series been used twice with success, and is the treatment of choice in cases which are not too extensive and where excision is not practicable. Some authors have reported on the use of X-ray therapy, and the general opinion seems to be that it is not very satisfactory. I have no experience of its use for these extensive lesions.

The treatment of choice is undoubtedly excision if it can be practised without producing a crippling deformity. In many the lesion is explored without a diagnosis being made, and in some it may be found possible to remove only part of the tumour which may have no clearly defined boundaries. Even then, however, excision of the main bulk of the tumour will be beneficial. Excision or partial excision was practised with complete success in 10 out of the 16 cases reported. The operation is often accompanied by less bleeding than would be expected, and in the case of a limb can be made almost bloodless by the use of a tourniquet.

Table I.—Summary of Cases

Case No.	Location	Age	Pain	Trauma	Rapid Enlargement	Skin Involvement	Phleboliths	Bone Involvement	Pre-operative Diagnosis	Treatment	Result
1	Masseter and facial muscles	5/12	No	No	No	Yes	No	No	Hæmangioma	Injection neovaricaine	Satisfactory
2	Infraspinatus	6	No	No	No	No	—	—	Lipoma	Excision	Satisfactory
3	Sternomastoid	23	No	No	No	No	—	—	Branchial cyst	Excision	Satisfactory
4	Entire lower limb	14	Yes	No	No	No	Yes	Yes	Hæmangioma	Nil	—
5	Trapezius	15	No	No	No	No	—	—	Neurofibroma	Excision	Satisfactory
6	Hand	18	Yes	No	No	Bluish patches	No	Yes	Hæmangioma	1. Diathermy 2. Radium	Unsatisfactory Satisfactory
7	Vastus intermedius	13	Yes	Yes	No	No	—	—	Lipoma	Excision	Satisfactory
8	Forearm (subcutaneous)	19	Yes	No	No	No	—	—	Hæmangioma	Excision	Satisfactory
9	Biceps and brachialis	12	Yes	No	No	No	No	No	Hæmangioma	Partial excision	Satisfactory
10	Gastrocnemius	10	No	No	No	No	No	No	Lipoma	Biopsy	Unchanged
11	Latissimus dorsi and erector spinæ	48	Yes	No	Yes	Yes	Yes	No	Hæmangioma	Partial excision	Satisfactory
12	Fingers	3	No	No	No	No	Yes	Yes	Hæmangioma	Radium	Satisfactory
13	Tibialis posterior	24	Yes	No	No	No	Yes	Yes	Hæmangioma	Partial excision	Unsatisfactory mass remained
14	(*a*) Platysma (*b*) Masseter and parotid gland	32	No	No	No	No	No	No	Hæmangioma Hæmangioma	Excision Partial excision	Satisfactory Satisfactory
15	Deltoid	26	Yes	Yes	No	No	Yes	No	Hæmangioma	Excision	Satisfactory
16	Psoas and kidney	19	No	No	No	No	No	No	Hæmangioma of kidney Muscle tumour not recognized	Nephrectomy Nil	Satisfactory Unchanged

SUMMARY

Sixteen cases of diffuse systemic angiomata are described in detail. The diagnosis is not difficult to make if the condition is remembered and resource made to radiology and aspiration. The methods of treatment are considered and excision is recommended whenever practicable.

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INTERDIGITAL SINUSES OF BARBERS' HANDS

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In 1880, in a paper on the postanal pilonidal sinus, Hodges wrote, "Hair-cutters are continually in trouble from the short hairs which penetrate the skin of their fingers and hands". Interest was not revived in lesions of barbers' hands until 1946 when Patey and Scarff described an interdigital sinus in a hairdresser, and later the same authors (1948) added 2 further cases to the literature. Up to date only 18 have been recorded, but both Patey and Scarff (1946) and King (1949) have remarked that the lesion is relatively common. The available relevant details of these published cases, many of which are described inadequately, are summarized in *Table I*.

Table I.—Clinical Summary of Recorded Cases

Author	No. of Cases	Sex and Age (years.)	Duration of Employment as Barber	Cutting Hand	Duration of Lesion	Interdigital Web Involved	Nature of Lesion
1. Allington, H. V., and Templeton, H. J. (1942) Templeton, H. J. (1942)	1	Male, 27	6 yr.	—	3–4 yr.	*a.* Index and middle R. *b.* Middle and ring R.	Multiple sinuses
2. Patey, D. H., and Scarff, R. W. (1946)	1	Male, 31	—	—	—	Ring and little R.	Sinus
3. Ewing, M. R. (1947)	1	Male, 52	Many years	—	6 wk	Middle and ring R.	Sinus
4. Patey, D. H., and Scarff, R. W. (1948)	2	*a.* Male, 34 *b.* Male, 53	20 yr. —	— —	3 mth. 4 yr.	*a.* Middle and ring R. *b.* Middle and ring L.	Sinus Sinus
5. Tait, G. B. (1948)	1	—	—	—	—	Middle and ring L.	Recurrent suppuration
6. Wilks, J. M. (1948)	1	—	—	R.	—	Thumb and index R.	Short sinus
7. Sames, C. P. (1948)	1	—	—	—	—	Middle and ring L.	Sinus
8. King, E. S. J. (1949)	4	*a.* Male, 72 *b.* Male, 42 *c.* Male, 37 *b.* Male, 53	Many years — — —	— — — —	4 yr. 2 yr. — —	Middle and ring L. ? Between middle and ring L. Middle and ring R. Index and middle L.	Sinus Shallow pit Pit Pit
9. Jenkins, D. H. (1951)	1	Male, 40	25 yr.	—	6 yr.	Index and middle R.	Sinus
10. Warren, C. M. (1951)	1	—	—	—	—	? Between index and middle R.	Multiple sinuses
11. Raw, S. C. (1951)	1	Male, 43	—	—	6 mth.	Index and middle (side not given)	Sinus distal to web
12. Downing, J. G. (1952)	1	Male, 71	40 yr.	—	Some weeks	Index and middle R.	Small red sinus
13. Hueston, J. T. (1952)	1	Male, 39	18 yr.	—	5 yr.	*a.* Index and middle R. *b.* Middle and ring R.	Single sinus Three openings
14. Waisman, M., and Olivetti, R. G. (1952)	1	Male, 49	—	—	3 mth.	Middle and ring L.	Sinus

Our attention was directed to this lesion when a hairdresser reported to the Surgical Out-patient Department with an interdigital sinus of the hand. We could find no trace in our records of a similar case having been treated in the hospital and were, therefore, surprised to find 4 others as a result of casual inquiry on our next visit to the hairdresser. We have since examined the hands of 77 gentlemen's hairdressers from eighteen different shops; of these, 10 showed the lesion in some stage of its development. We have also inspected the hands of 61 ladies' hairdressers and have not found one lesion of this type. Waisman and Olivetti (1952) have rightly made the point that "to many of the victims such a relatively trivial and non-disabling occupational event hardly merits seeking medical advice".

CLINICAL FEATURES

The details of our cases are summarized in *Table II*.

DEVELOPMENT OF LESION

Gross Features.—Hairs tend to collect in the webs of the fingers of barbers during the course of their day's work and not infrequently the short, sharp hairs from the heads of male customers may penetrate the epidermis and project from the skin surface (*Fig.* 334). As a result of secondary infection in the superficial corium small pits are formed, just dorsal to the web margin (*Fig.* 334), and more hairs gather in them, with consequent persistence and

aggravation of these reactive changes. Small sinuses are produced (*Fig.* 335) and occasionally multiple ones develop in the same interdigital cleft (*Fig.* 336). With the entry of more hairs through the sinus into the deeper layers of the corium and after further infective episodes, a longer sinus results. An abscess may form at its tip and point in the palm or dorsum of the hand (*Fig.* 337), and it may rupture or be incised to produce a tunnel or fistula (*Figs.* 338, 339). The lesion is frequently present, and although there may be superadded acute inflammation and suppuration little destruction of the new epithelium has been seen (*Figs.* 341, 343). The sinus may burrow deeply in the corium (*Fig.* 344) or even in the subcutaneous tissue, and superficially its epithelial lining is supported by dense connective tissue in which hair shafts may be embedded. Hair shafts, however, are usually present not only within the sinus but also in

Table II.—Clinical Summary of Authors' Cases

Case	Sex	Age	Duration of Employment as Barber	Cutting Hand	Duration of Lesion	Interdigital Web Involved	Nature of Lesion	Treatment
1. J. B.	Male	42 yr.	25 yr.	R.	2 yr.	*a*. Thumb and index R. *b*. Index and middle L. *c*. Index and middle R.	Shallow pits (*Fig.* 334) Multiple sinuses (*Fig.* 336). Scar	Under observation Excision
2. R. Y.	Male	36 yr.	20 yr.	R.	Not noticed before examination	Index and middle R.	Three shallow pits, one with hairs sticking into it (as *Fig.* 334)	Under observation
3. J. H.	Male	42 yr.	25 yr.	R.	1 yr.	Index and middle R.	Early sinus. No hairs obvious (*Fig.* 335). Had two acute suppurative episodes	Excision
4. W. E.	Male	30 yr.	18 yr.*	R.	1 yr.	Thumb and index L.	Early sinus. No hairs (as *Fig.* 335)	Excision
5. A. B.	Male	50 yr.	40 yr.*	R.	6 yr.	Index and middle R.	Early sinus (as *Fig.* 335)	Refused operation
6. T. C.	Male	46 yr.	25 yr.	R.	2 yr.	Index and middle L.	Short quiescent sinus with hairs. Previous acute suppurative episode	Under observation
7. P. McG.	Male	24 yr.	10 yr.	R.	5 days?	Index and middle L.	Multiple infected sinuses. Hairs present in two (as *Fig.* 336)	Excision
8. M. B.	Male	46 yr.	20 yr.	R.	2 mth.	Middle and ring R.	Sinus with abscess pointing in dorsum of hand (*Fig.* 337)	Excision
9. T. McC.	Male	68 yr.	52 yr.	R.	25 yr.	Index and middle R.	Tunnel through to palm of hand (*Fig.* 338). History of several suppurative episodes	Refused operation
10. R. E.	Male	67 yr.	57 yr.*	R.	15 yr.	Index and middle R.	Tunnel through to dorsum of hand filled with hairs (*Fig.* 339)	Excision
11. J. R.	Male	56 yr.	44 yr.*	R.	Some years. Now healed	Index and middle L.	Fine scar	Nil

* It will be noted that these patients commenced work at the early ages of 10 and 12 years. This is explained by part-time work in family businesses.

hairs from the lesion illustrated in *Fig.* 339 were all cut hairs; they varied in degree of pigmentation and in thickness (*Fig.* 340), and were obviously derived from a number of different heads. The presence of protruding hairs is not, however, a constant feature (*Figs.* 335, 337, 338), since before reporting for medical attention some of the barbers take pains to remove all the obvious hairs from the sinus opening.

Microscopical Features.—Of the 6 lesions excised 4, including 2 cases with multiple sinuses, were examined in serial section and when necessary consecutive sections were studied. Although the two oldest lesions were not serialized, numerous sections at different levels were examined.

The very early lesions have not been studied histologically as excision at this stage is not justifiable. In all the sinuses the lining consists of non-pigmented squamous epithelium which does not possess rete pegs (*Figs.* 341, 342). At the sinus tip a granulomatous the granuloma at its tip (*Fig.* 345). If the further entry of hairs is prevented, and if the infective process is controlled, the sinus tip may close. It should be emphasized that examination of serial sections is frequently essential to establish whether or not a sinus is completely closed, for its opening may be very small. In the tunnel epithelialization is complete (*Fig.* 346), but there is evidence of chronic non-specific inflammation around it. The development of the lesion is illustrated diagrammatically in *Fig.* 347.

Arising out of more detailed study of the material, two points merit emphasis. First, a sweat-duct is present in the epithelium lining the tunnel (*Figs.* 346, 348) and in several closed pits of varying depth one was found winding through the epithelium of the base. Secondly, the *cellular reactions to the hair shafts*, which differ in thickness and in degree of pigmentation, are variable. In some cases numerous polymorphs are present around the hair shafts, while

in others lymphocytes and plasma cells predominate (*Fig.* 349). Foreign-body giant-cell reactions are frequently seen (*Fig.* 350) and sometimes the hair shafts are surrounded by young connective tissue (*Fig.* 351). When the hairs have been present in the tissues for a longer period, dense connective tissues encircles them (*Fig.* 351), and at times they may be seen embedded in the fibrous tissue supporting

FIG. 334.—*Case* 1. A short hair is sticking into and projecting from the skin of the web between the thumb and index finger of the right hand. Two shallow pits are present in the skin near it.

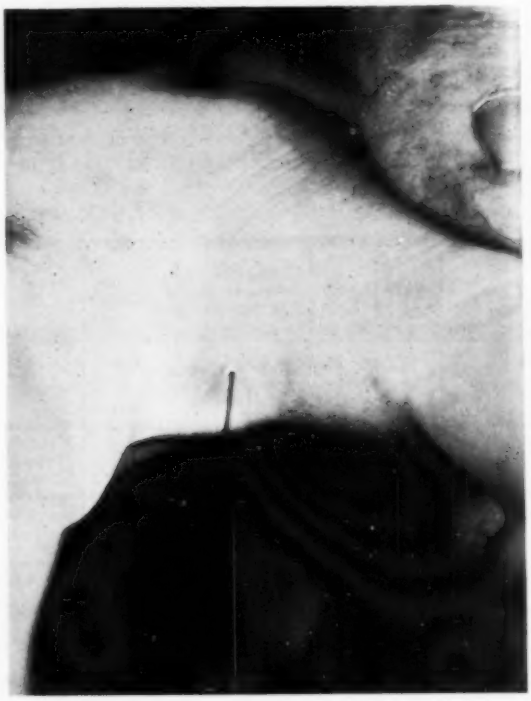

FIG. 335.—*Case* 3. A probe, which passed for a distance of 1 mm., is present in an early sinus, the opening of which is just dorsal to the web margin.

FIG. 336.—*Case* 1. Multiple interdigital sinuses.

FIG. 337.—*Case* 8. There is a large sinus opening dorsal to the web margin and an abscess is pointing in the dorsum of the hand.

the superficial part of a sinus. Occasionally hairs are present at a considerable distance from the epithelial lining of the sinus, frequently in association with small blood-vessels. No hairs were seen in the connective tissue supporting the epithelium of the tunnel examined. In a few instances, irregularity of the hair outline has been observed (*Fig.* 352) and some appear to be in the process of absorption. Patey and Scarff (1946) described a hair follicle in the wall of the sinus of the first case they reported. Hair follicles, however, are not present in the skin of the finger webs and we have not seen one in the wall of any of the lesions.

FIG. 338.—*Case* 9. A probe is present in a tunnel which has emerged in the palm of the hand.

FIG. 339.—*Case* 10. A sheaf of hairs in a tunnel which has emerged in the dorsum of the hand.

DISCUSSION

Factors determining the Entry of Hair and the Site of the Lesion.—Barbers are well acquainted with the traumatic and infective properties of short clipped hairs from male heads. These hairs may stick into the skin covering many parts of the body—the finger webs, under the nails, in the nail beds, the trunk, the legs, and feet; they may also penetrate the conjunctiva (Warren, 1951) or may be swallowed. A small inflammatory lesion of the skin may be produced around the hairs, but once they are removed the infection subsides.

It appears, however, that the only frequent site of formation of a chronic lesion is in the skin of the finger webs. Any of these in either hand may be involved, but the sinus occurs most commonly between the forefinger and middle fingers and between the middle and ring fingers of the right hand. Hairs tend to collect in the finger webs of barbers' hands during the course of their work and several factors may be responsible for this. Tait (1948) has pointed out that when a barber runs his hands through a customer's hair the clipped hairs gather in the webs.

It is also clear that hairs will accumulate more readily in a cleft, particularly if, as in this case, it may be moist and sticky with soap, hair creams, and sweat. It is, therefore, likely that there is always a greater concentration of hairs in the finger webs than on any other part of the body surface.

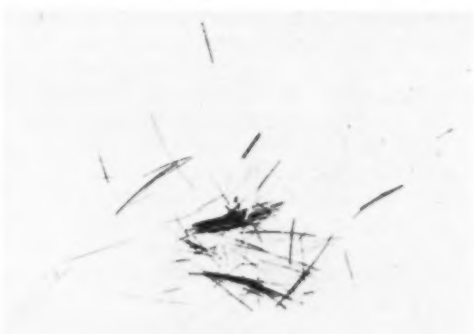

FIG. 340.—*Case* 10. Hairs of varying thickness and colour removed from the lesion illustrated in *Fig.* 339.

Numerous theories have been suggested to account for the entry of hair into the skin. Sames (1948) has offered the explanation that the pressure of the comb on the finger webs may be the causal force; suction as a result of production of negative pressures has been postulated by Patey and Scarff (1946, 1948), while Ewing's patient implicated shampooing (1947). We believe, however, that inadequate stress has been

laid on the ability of short, sharp, clipped hairs to penetrate the epidermis in any situation, on the concentration of those hairs in the finger webs, and on the fine texture of the skin at this site. Matheson (1951) in his report of an interdigital sinus in a sheep-shearer, where the traumatic agent was wool fibre,

FIG. 341.—*Case* 7. Early lesion showing epidermis on either side of sinus opening with acute inflammation at tip. Hæmalum and eosin. (× 44.)

suggested that a particular type of movement by adjacent fingers in full adduction may act as a propelling force. After shampooing, barbers often run the fingers of one hand through the interdigital clefts of the other to remove the soap adhering there and we think that this action may be implicated at times. It would appear that these several movements, actions, and forces are not essential factors for determining the initial entry of hair, and similarly we do not consider that pre-existing creases or pits in the skin need necessarily be present as inferred by King (1949) and Waisman and Olivetti (1952). It is of interest, in this connexion, that the second case reported by Patey and Scarff in 1948 gave a definite history of an accidental scratch with wire in an interdigital web immediately before a sinus developed.

As a result of penetration of the epidermis by a hair or hairs, secondary infection and foreign-body giant cells are stimulated and small pits are formed. More hairs readily accumulate in these and the process continues with sinus formation. The entry of more hairs into the corium through the sinus opening may perhaps be aided by suction forces.

Pathology.—The sinuses in our series were all lined by squamous epithelium and even in those cases with inflammation and suppuration at the sinus tip there was little destruction of it. The epithelium grows in a very regular fashion into the granulation tissue which precedes it, and in some cases the spinous cells appear to be in advance of the basal cells. This process continues as long as hairs are carried along the sinus to be lodged in the corium or subcutaneous tissue at its tip. If the entry of further hairs is prevented and the infection subsides the opening at the sinus tip may become lined by epithelium to form a closed pit. Hairs may accumulate readily in a pit of this nature and, with penetration of the new epithelium by hairs and consequent re-infection, the lesion progresses and a tunnel is eventually produced (*Fig.* 347). In the tunnel (*Case* 10) there was evidence of chronic inflammation in the fibrous tissue around the epithelium and on general principles it seems reasonable to suggest that a squamous-cell carcinoma might develop in such a lesion as a result of long-standing chronic irritation, although no such case has been recorded.

The variable *cellular reactions to the hairs* in the corium have been described and it has been stated that some of the hairs appear to be in the process of absorption. That eventual absorption of hair may occur is supported by the fact that none were present in the connective tissue surrounding the epithelium lining the fistula, which was the oldest lesion examined. On the other hand, hairs may be very

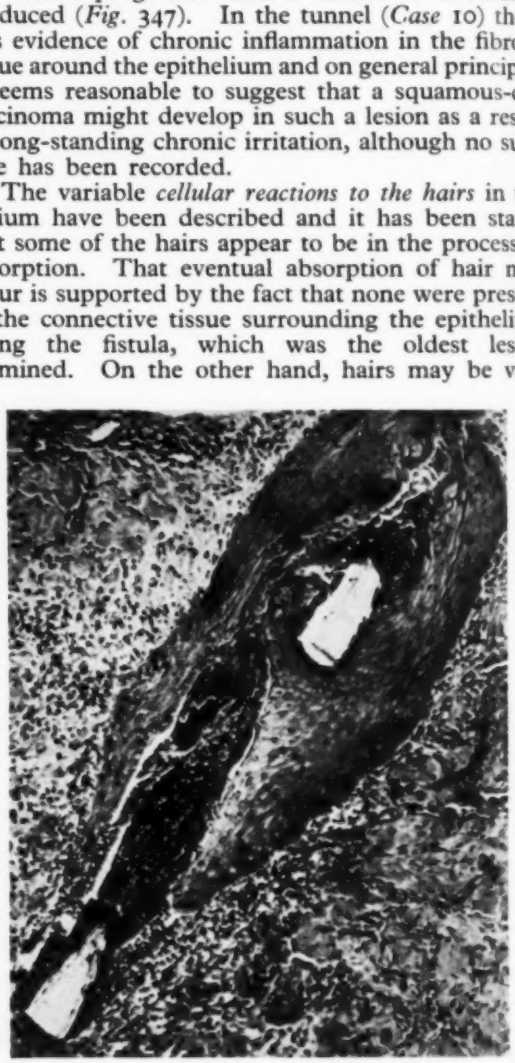

FIG. 342.—*Case* 7. A hair is present within the sinus and also at its tip where there is a granulomatous reaction. Hæmalum and eosin. (× 76.)

few in number or even occasionally absent in an active lesion, and Peacock (1953) claims that hairs introduced into the skin of experimental animals are not altered after several years. During the preparation of the material for microscopical examination some hairs become dislodged and, further, as the degree of pigmentation and thickness varies from hair to hair it is difficult to assess with any degree of certainty the changes produced in them

It is clear that further investigation of the fate of hairs in the tissues is essential before one is entitled to make a dogmatic statement as to whether or not absorption may occur.

The presence and apparent *reformation of sweat-ducts* in the epithelium lining the bases of the closed pits and the fistula have already been mentioned. Some of these pits are relatively shallow and it is possible that the epithelium lining them and the sweat-ducts within it are part of pre-existing structures which have been deformed as a result of cicatrization deep to them. This explanation, however, does not account for the presence of a sweat-duct in the epithelium lining the base of pits deeply situated in the corium or for the sweat-duct in the epithelium lining the tunnel and, therefore, one must apparently accept the fact that sweat-ducts have re-formed in new epithelium.

Relationship to Other Hair-bearing Lesions.—Clearly the barber's sinus is different from other hair-bearing lesions in that the hairs are of extraneous origin. Further, it differs from most of them, including the postanal sinus, as the hairs are primarily responsible for its development. We would emphasize that this lesion is produced by the short, sharp, clipped hairs from male heads and that we have not seen or heard of a case in a ladies' hairdresser who is exposed, in the main, to longer hairs.

Pinkus (1943) has written an excellent description of "Chronic Scarring Pseudofolliculitis of the Negro Beard", the clinical features of which had been published previously by Greenbaum (1935). It is a papular, sometimes pustular, eruption affecting the hair-bearing part of the skin of the face and neck in adult male negroes and is well developed in more than 10 per cent of them. The lesion is caused by the shaved hairs curling back into and penetrating the epidermis. This is an example of a lesion directly due to the host's hair, and again the trauma is caused by the sharp cut end.

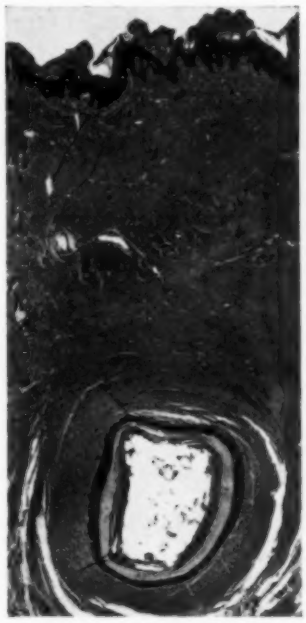

FIG. 344.—*Case* 8. Transverse section of superficial proximal part of sinus. The sinus is deeply placed in the corium below the level of the sweat-glands. It is lined by keratinizing squamous epithelium which is supported by dense connective tissue. Hæmalum and eosin. (× 12.)

FIG. 345.—*Case* 8. Abscess cavity with wall of granulation tissue in which some hairs are embedded. A few hairs have been dislodged during the preparation of the section, leaving empty spaces. Hæmalum and eosin. (× 10.)

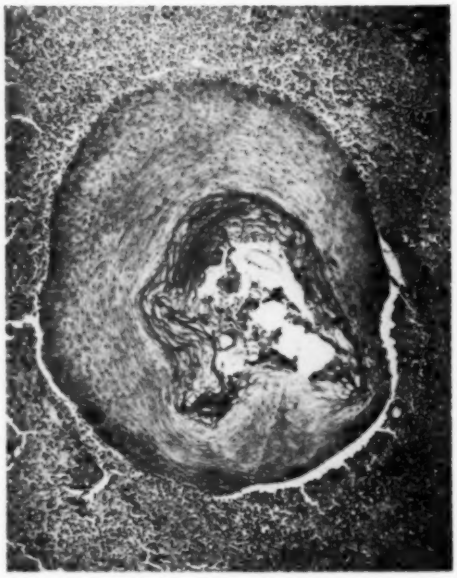

FIG. 343.—*Case* 8. Transverse section of sinus tip. The sinus is lined by squamous epithelium and there is no keratin formation. Two or three hairs are present within the sinus, and around it there is granulation tissue with acute inflammation in places. Hæmalum and eosin. (× 40.)

Patey and Scarff's papers (1946, 1948) on the relationship of this lesion of barbers to the postanal pilonidal sinus stimulated much writing in the medical press. In view of their experience of the barbers' lesion, combined with a study of 21 postanal sinuses,

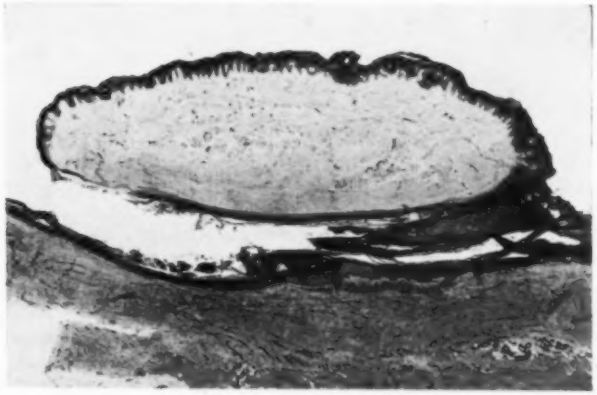

FIG. 346.—*Case* 10. A completely epithelialized tunnel. There is some chronic inflammation deep to it. Hæmalum and eosin. (× 8.)

they suggested that the great majority of postanal pilonidal sinuses were acquired and not congenital, and that hairs and suction forces were important factors in their production. The actual *modus operandi*

of the hairs was not certain. It was suggested, however, that the hairs of the postanal region were the agents primarily responsible for the initial stage in the development of a sinus in this situation. Later Patey (1952) pointed out that although the

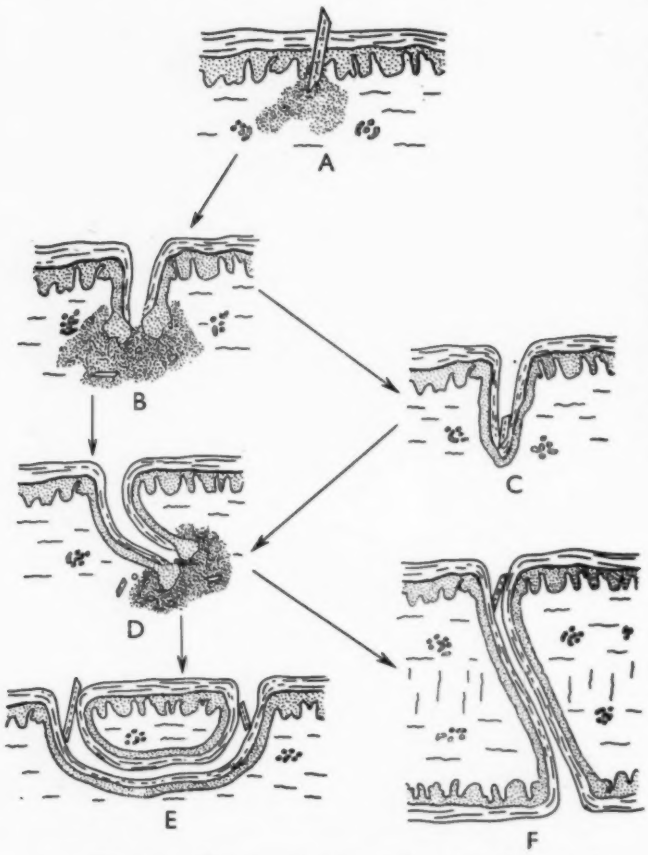

FIG. 347.—*Diagrammatic representation of development of lesion.* A, Hair penetrating epidermis with inflammatory reaction around it in superficial corium. B, Early sinus. C, A closed pit. Infection subsided. D, Longer sinus developing from B or C. E, Tunnel emerging in dorsum of hand. F, Tunnel emerging in palm of hand.

postanal sinus might originate in a pilosebaceous follicle, which King (1947) had previously suggested, the penetration of the epidermis by hair was not the original cause of the infection. In our view it is most unlikely that the free end of the long hairs of the postanal region could penetrate the epidermis. It will be recalled that we have not seen or heard of a case of interdigital sinus of the hand in a ladies' hairdresser, who is exposed, of course, to longer hairs in the course of her employment than her male counterpart. Patey also suggested that the chronicity of the postanal sinus is explained by the constant sucking in of hairs and debris once a break has been produced in the skin surface.

There are, however, many so-called postanal pilonidal sinuses which do not contain hairs and in Patey and Scarff's (1946) series 11 out of 21 lesions fell into this category. We have already stated that the ultimate fate of hairs in the tissues is not known, and although we have suggested that absorption may occur, we believe that it must take some considerable time. Therefore, if hairs are as important in the development of the postanal lesion as Patey and Scarff have claimed, one would expect to find a higher percentage of hair-bearing sinuses. Wilson (1946), Newell (1946), Ewing (1947), and Sames (1948) were not in agreement with the views of Patey and Scarff, but Downing (1952) supported them.

Nomenclature.—Many of the previous reports on interdigital sinuses of barbers' hands have referred to them as ' pilonidal sinuses '. This term has been used recently, and in our view often incorrectly, to describe a number of lesions differing in aetiology and pathology. It was first applied by Hodges (1880) to the postanal sinus, and since then pilonidal sinuses have been reported in the perineum (Smith, 1948), the axilla (Aird, 1952), and the suprapubic region (MacLeod, 1953). Aird (1952) claimed that the axilla was the sixth site of pilonidal suppuration to be described, the other five being in the postanal region, the perineum, barbers' hands, the sole of the foot, and the umbilicus. We are of the opinion that the term ' pilonidal sinus ', which is purely descriptive, should not be used at least without qualification, when the pathology and mode of development of the lesion is known. Further, it is surely illogical and

FIG. 348.—*Case* 10. Higher magnification of part of *Fig.* 346, to show sweat-duct in epithelium lining tunnel. Hæmalum and eosin. (× 64.)

wrong to apply it when there is, on the one hand, no sinus, or, on the other, no hair present in it. Similarly, a suppurative lesion in a hirsute area does not necessarily justify the use of the label ' pilonidal '. We suggest, therefore, that the term ' pilonidal sinus ' be retained only for those cases of postanal sinus which are not of a congenital nature and which contain hairs and, perhaps, secondly, for the perineal lesions described by Smith (1948). We hold the view that it should not be used to name the lesions described in the other five sites—the umbilicus, the sole of the foot, the axilla, the suprapubic region, and the sinuses of barbers' hands—as it is either a misuse of the term or misleading and confusing.

A certain Dr. Jackson is reputed to have described the first umbilical case at a meeting of the Boston Society for Medical Improvement after a paper had been read by Warren (1854) on the postanal lesion. In the records of the Society we find that " Dr. Jackson referred to a specimen, in the Society's cabinet, of a lock or pellet of hair taken from near the umbilicus ", but there is no mention of an inflammatory lesion or of sinus formation. Inflammatory lesions of the umbilicus were, however, described by Hodges (1880), but apparently sinuses did not develop. Aird (1952) has seen recurrent umbilical suppuration in a hirsute, young, female patient but she refused operation and consequently histological examination of the lesion was not possible. In our view, therefore, there is insufficient evidence for including the umbilicus as one of the sites of pilonidal sinus formation or of pilonidal suppuration.

In 1880 Hodges pointed out that a woman's hair or horsehair from a mattress " may insinuate itself beneath the skin " of the sole of the foot if trod upon on the floor. He also records that Jackson had exhibited to the Boston Society for Medical Improvement a bristle which had been extracted from a foot, " into which it had penetrated unconsciously to the patient for a distance of one and a third inches ". If we accept these as pilonidal sinuses of foot, and we have failed to find any other references in the literature, then the sites of formation are manifold as hairs may at times stick into the skin of a barber on any part of the body surface.

Aird's case of ' pilonidal sinus ' of the axilla was not thus diagnosed until after the third excision of a persistent sinus. On microscopical examination of this third specimen two hairs were seen, though none had been detected in the material removed at the first two operations. As a result of biopsy or of incision of the skin, particularly in a hirsute area, hair shafts may be dislodged and embedded in the corium. It is possible that this may be the explanation of their presence in the third specimen and we consider that in the first instance it did not satisfy the conditions to justify the diagnosis of pilonidal sinus.

MacLeod (1953) clearly described his case as a ruptured, infected dermoid cyst of the suprapubic

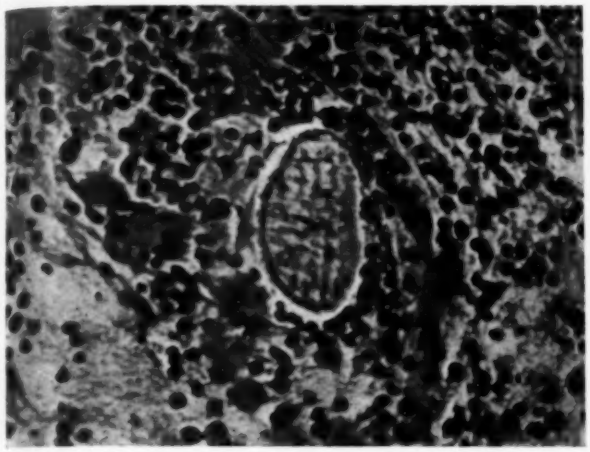

FIG. 349. – *Case* 8. Hair shaft surrounded by inflammatory cells, mainly lymphocytic in nature. A few macrophages are seen to the left. Hæmalum and van Gieson. (× 293.)

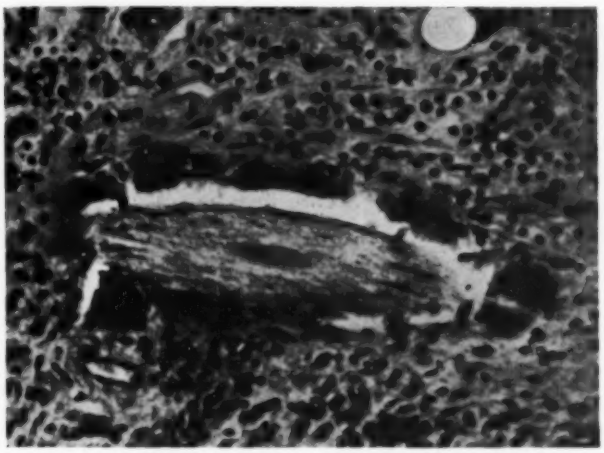

FIG. 350.—*Case* 8. Hair shaft surrounded by multinucleate giant cells. Hæmalum and eosin. (× 220.)

FIG. 351. – *Case* 8. The hair shaft to the left is surrounded by young connective tissue, while the one to the right is encircled by dense connective tissue. Hæmalum and eosin. (× 165.)

FIG. 352.—*Case* 8. There is irregularity of the hair shaft. Hæmalum and van Gieson. (× 154.)

region, but puts it on record as the seventh site of pilonidal sinus formation and suggests that pilonidal suppuration could occur in a dermoid cyst of the skin in any situation. We have recently seen one on the nose but we prefer to call it a ruptured, infected dermoid cyst than to claim it as the eighth site of pilonidal sinus formation. We have already criticized the use of this purely descriptive label when the lesion is known to be a ruptured dermoid cyst (Currie, Gibson, and Goodall, 1953).

Finally, we consider that the lesions which form the basis of this communication should be known as ' interdigital sinuses of barbers' hands ' and that the confusing term ' pilonidal ' should be avoided.

PREVENTION AND TREATMENT

There is little doubt that if barbers paid sufficient attention to hand hygiene and were careful to remove all the hairs from their finger webs at the end of each day's work this lesion could be prevented. It is our opinion that this should be brought to the notice of all barbers as the condition is common and, although not of a severe nature, is an irritating disability.

In the early shallow pitted stage of the lesion (*Fig.* 334) excision has not been advised or undertaken as, if the patient removes all hairs from the web each night, it does not progress. Once it has advanced beyond this stage, however, surgical excision is the treatment of choice, particularly if repeated infective episodes have occurred, and care must be taken to remove any deeply situated granuloma.

Six of the eight patients so advised accepted operation. Two with multiple sinuses required a free skin-graft after excision, the remainder being closed by direct suture. Primary healing occurred in each case and the results have been satisfactory.

SUMMARY

1. The literature of interdigital sinuses of barbers' hands is reviewed.

2. Eleven cases are described bringing the total in the literature to 29. This number bears no relation to the incidence of the lesion as it is common among barbers.

3. The pathology and mode of development are described and the tissue reaction to hairs is discussed.

4. The nomenclature of hair-bearing lesions is considered and the indiscriminate use of the term ' pilonidal ' is deprecated. The ' pilonidal sinuses ' described in the axilla, the umbilicus, the sole of the foot, and the suprapubic region are not accepted as such. The lesions which form the basis of this communication should be known as ' interdigital sinuses of barbers' hands ' and the confusing term ' pilonidal ' should be avoided.

5. The treatment of the lesion is complete excision and prevention is possible with careful hygiene.

It is a pleasure to acknowledge our indebtedness to Professor G. L. Montgomery for his interest and encouragement; to Mr. P. J. Elliott for the photography; to Mr. R. W. Chalmers of the Stow College of Hairdressing for his help; and to the hairdressers in the region for their co-operation.

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PERFORATION OF THE SIGMOID COLON

By PHILIP GOATCHER

LATE SURGICAL REGISTRAR, COUNTY HOSPITAL, WHISTON

" The chief defect of Henry King
Was chewing little bits of string.
At last he swallowed some which tied
Itself in ugly knots inside."—H. Belloc.

SWALLOWED indigestible foreign bodies are a common enough finding in the gastro-intestinal tract. The great majority of these pass uneventfully, but if they are sharp a perforation may result at any point in their course, some sites being more vulnerable than others.

Carp (1927) estimated that more than 80 per cent of such bodies passed harmlessly, while less than 20 per cent became lodged in some part of the gastro-intestinal tract and were therefore liable, sooner or later, to produce a perforation. When

the foreign body is neither sharp nor hard this risk is indeed small.

In the case here reported the patient, over a period of years, had repeatedly swallowed small pieces of cloth which, accumulating in the redundant sigmoid colon, produced a palpable mass, mild signs and symptoms of intestinal obstruction, and finally perforated the colon at the place of impaction.

CASE REPORT

Mrs. E. L., a 45-years-old housewife, entered hospital on Dec. 15, 1951. She complained of abdominal pain and constipation, which was unrelieved by an enema ordered by her private doctor. One week prior to admission she had some diarrhœa and on the day of admission had retched a little.

Three years previously the patient had suffered similar symptoms which had passed off after a few days in bed.

PHYSICAL EXAMINATION.—The patient was well-nourished, well-preserved, and a healthy-looking middle-aged woman. The temperature was 99·4° and pulse 86. Her tongue was moist and furred.

The abdomen was not distended. There was a slightly tender tumour in the left iliac fossa, extending from the pelvis upwards towards the splenic flexure. It felt like a colon loaded with fæces. Rectal examination showed the rectum to be empty. The mass was readily palpated through the rectal wall, while bimanual examination proved it to be mobile.

An enema produced copious flatus, but no fæces.

A diagnosis of incomplete large-bowel obstruction was made and appropriate investigations ordered.

Forty-eight hours later the patient complained of sudden severe abdominal pain and was found to be collapsed and pale with a subnormal temperature, rapid pulse, and rigid tender abdomen. From this clinical picture a diagnosis of peritonitis, due to a perforation of large bowel, was made.

After the usual restorative measures had been used to improve her general condition it was decided to carry out a laparotomy.

Through a left hypogastric paramedian incision the abdomen was explored. There was a little fæcal fluid in the left iliac fossa and the sigmoid was rigid from œdema and thickened over a length of some 3 in. (8 cm.). A perforation approximately ¼ × ⅛ in. (1 × 0·5 cm.) in dimension was found in that area, and the proximal sigmoid and ascending colon seemed to contain firm fæces.

FIG. 353.—Operative specimen, showing the excised piece of sigmoid colon, the bolus of cloth, and the perforation.

FIG. 354.—The pieces of linen and cotton material which produced the perforation

AT OPERATION.—General anæsthetic (by Dr. H. T. Kay)—pentothal, gas, oxygen, ether, and flaxedil.

The appearance and mobility of the involved area of colon precluded diverticulitis, while the lack of true hardness and the absence of adjacent lymph-gland enlargement made a diagnosis of carcinoma unlikely.

A double-barrel was made from the proximal and distal ends, the affected loop exteriorized and a Paul-Mikulicz resection performed. Some 6 in. (15 cm.) of large intestine was excised and an enterotome applied. The peritoneal cavity was sucked out and the abdomen closed without drainage.

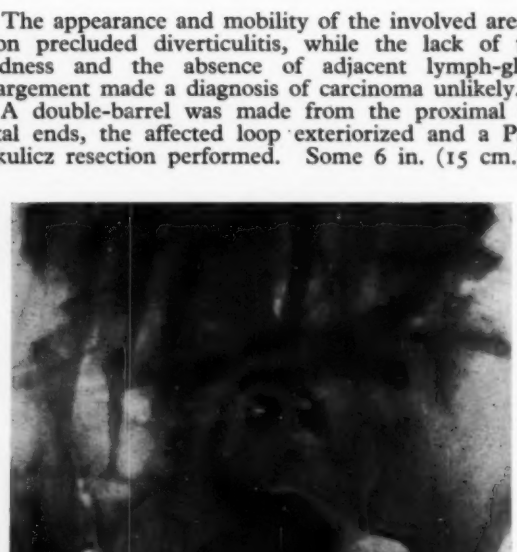

FIG. 355.—Radiograph of barium enema taken after the resection and colostomy to demonstrate the approximate site of the perforation.

SPECIMEN.—When this was opened I was surprised to find a mass comprised of pieces of cloth, forming a bolus of such a diameter as to fill the lumen of the incised piece of sigmoid. Closer examination proved this to consist of sixty pieces of heavy cotton and linen fabric varying in width from ½ to 2 in. (1 to 5 cm.) and 2 to 6 in (5 to 15 cm.) in length. (*Figs.* 353, 354.)

PROGRESS.—Intravenous saline, gastric suction, and intramuscular streptomycin were continued and after 24 hours of pyrexia the patient's general condition had improved sufficiently to discontinue the drip and suction.

The colostomy functioned two days later, when more cloth was extruded. Pieces of cloth continued to be passed for a further ten days, by which time 90 g. had been collected. In all there were 250 pieces of material, with an area estimated at quite 600 sq. in. (3750 sq. cm.). In addition there was a mass of linen thread.

Later a barium enema was given to demonstrate the site of the colostomy and so to estimate the position of impaction (*Fig.* 355).

The patient was not observed to eat cloth while in hospital, but when questioned she reluctantly admitted chewing and swallowing pieces of material at home. She was in all other ways an apparently normal person who was capable of running a home and caring for a son of 15 years of age. It embarrassed her to discuss her strange practice. Her husband (a policeman) stated that he had for years tried to dissuade her from chewing pieces of cotton and linen.

When the incision had healed and the patient fully recovered, the question of whether to close the colostomy gave rise to the problem of a possible recurrence. The advice of a psychiatrist (Dr. H. Astley Cooper) was sought. After two sessions with the patient he advised psychiatric treatment, but considered that closure could be safely undertaken.

After a period of four weeks at home the colostomy was closed by the extraperitoneal method. The patient was delighted to have this done and was discharged fourteen days later with normal bowel habits.

DISCUSSION

The literature abounds in records of innumerable foreign bodies of infinite variety found in the intestinal tract. They form a bizarre collection rivalled, it seems, only by those introduced into other bodily orifices.

Foodstuffs.—Frequently foodstuffs have been incriminated as the cause of intestinal obstruction and less often of perforation. Ward-McQuaid (1950) refers to the now well-recognized condition of impacted residue of ingested food. The incidence may be gauged from his investigation of 875 cases of obstruction from causes other than herniæ. In his series 50 cases were due to foreign bodies and 5 of these resulted from food.

The commonest site of impaction was the terminal ileum and the commonest causes were persimmons and peaches, but oranges, apples, cherries, grapes, apricots, pears, gooseberries, coconut, almonds, beans, oats, cucumber, poppy seeds, and even grasshoppers were listed as causes in that review of the literature.

Foreign Bodies.—Long as the above list is, it is eclipsed by the number and variety of foreign bodies other than foodstuffs, a list of which would seem to be almost without limit. Pins, nails, screws, hairpins and slides, needles, artificial dentures, toothpicks, pieces of metal and wood, bones of various animals, a porcupine quill, and the foot of a lark are but a few of those referred to in the medical literature.

To these Weinstein, Roberts, and Sasse (1950) have added 1 case of obstruction produced by a condom filled with water and refer to one other similar reported case. On both occasions the offending object was believed to have been swallowed by the patients, who were men.

Gall-stones, fæcoliths, and impacted fæces seem to be the materials actually produced in the body which are capable of causing obstruction and possibly perforation. Gall-stones most often become arrested in the terminal ileum, although Lloyd Williamson (1952) reports a case of obstruction with perforation caused by a gall-stone becoming impacted in the sigmoid colon, the same site as in the case reported here.

Sites of Perforation. — Macmanus (1941) analysed 93 cases of perforation in the intestine caused by ingested foreign bodies. The striking feature of his findings is the great predominance of perforations at the lower ileal and cæcal regions (73 per cent). One may expect the arrest of a foreign body to have occurred at one of the relatively fixed and narrowed points in the intestinal tract and the perforation at or just proximal to one such point. Such sites would be the cardio-œsophageal junction, the pylorus, and the angles of the first and second parts of the duodenum. Such is not the case.

Sometimes the exact site of perforation cannot be identified since the object is found lying in an abscess cavity in one of the various recesses of the peritoneal cavity.

In 83 cases Macmanus found the sites of perforation to be as follows: Appendix 33 cases, ileum 10, cæcum 10, Meckel's diverticulum 10, sigmoid 5, hepatic flexure 2, duodenum 1, transverse colon 1, and descending colon 1. These findings indicate that the sigmoid colon is not the least common site of arrest of an ingested foreign body.

Perforation in Disease.—Clinical experience teaches that in any person over the age of 40 years, with an antecedent history of lower abdominal discomfort, who presents a febrile course and a picture of diffuse peritonitis, the cause is frequently a complication of diverticulitis, and the problem is how to treat it. In this case the patient was 45 and the previous history led one to expect some underlying disease of the bowel.

In a series of 1400 autopsies, Weinberger (1945) found 19 cases of perforation of the large intestine. Of these, 10 were due to diverticulitis and 6 were associated with carcinoma. It is reasonable to assume that when a perforation presents, these two conditions must be suspected, diverticulitis being more common than carcinoma.

Other diseases are usually diagnosed before the perforation occurs, so that the surgeon is less likely to be confronted for the first time with a patient suffering from peritonitis as a complication of one of these. Such diseases are advanced ulcerative colitis, dysentery (amœbic and bacillary), and tuberculosis. Simple and stercoral ulcerations with perforation are also rare, as are perforations due to to salmonella infections and *Ascaris lumbricoides* infestations.

Hawe (1951) reported a case of perforation occurring in the upper third of the rectum above a rectal stricture due to lymphogranuloma venereum.

Mechanical Causes.—Direct or indirect trauma may result in a tear through the full thickness of the colon wall. Such trauma may arise either outside or within the lumen of the intestine.

Penetrating injuries, which are common enough in battle casualties, obviously can and do produce perforations. Less obvious is the crushing injury to the abdomen which may also cause contusion, tearing, or a bursting of the intestinal wall. As the colon is both muscular and distensible, considerable force is required to produce this result.

Acute non-malignant perforations of the colon may be produced by forces acting within the gut and these similarly require considerable force. The introduction of compressed air, colonic washouts, sigmoidoscopies, and even enemata may produce a tear in the wall of an apparently normal colon.

It would seem that some obstruction is necessary to make possible rupture by a too vigorously administered enema, for the gradual distensibility of the entire colon is enormous.

Burt (1931) calculated from experimental data that a pressure of approximately 5 lb. per square inch is required to rupture the sigmoid. Yet Lataix (1949) was able to report 2 cases of spontaneous perforation. In the case reported here the perforation was not a tear but a necrosis due to pressure from the mass of cloth, with possibly superadded infection. This was the opinion of the pathologist as well as myself and the patient had not been given an enema for many hours prior to the sudden development of peritonitis.

COMMENT

This adds one more unusual and interesting case to the long list of reports of ingested foreign bodies and their complications.

The following features are unusual:—

a. It is remarkable that a foreign body which is neither sharp nor hard should produce a perforation of the colon. In most recorded cases the offending agent was sharp or at least composed of a hard material.

b. One would expect a case of perforation produced by such material to be associated with or be preceded by well-defined signs and symptoms of intestinal obstruction. This was not so.

c. Although a not uncommon finding, it is remarkable that a foreign body (in this case pieces of cloth) can traverse the pylorus, duodenum, and ileocæcal regions to impact in the redundant sigmoid colon.

It has been the experience of some authors that the prognosis in large gut perforations produced by foreign bodies is good, particularly with the aid of antibiotics. This proved so in this case.

SUMMARY

A case of ingested cloth (some 3700 sq. cm.) producing a perforation of the sigmoid colon with minimal obstructive signs and symptoms is described.

Literature on the subject of the aetiology of obstruction and perforation of the intestine is reviewed from the following aspects: (*a*) Foodstuffs, (*b*) Foreign bodies, (*c*) Sites of perforation, (*d*) Perforation in disease, (*e*) Mechanical perforation.

I wish to thank Mr. R. W. Doyle for permission to publish this case report and Dr. W. K. McGinley, pathologist, for his co-operation.

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INTRA-ABDOMINAL TORSION OF AN APPENDIX EPIPLOICA

By W. MURDIE, Bedford

Much interest has recently been aroused in the pathology of the appendices epiploicæ. They have been shown to undergo torsion; initiate intussusception (Harte, 1943); cause intestinal obstruction (Klingenstein, 1924); simulate a strangulated inguinal hernia (Adler, 1908); and, even in their normal state, to be mistaken for a normal vermiform appendix (Latimer and Hinshaw, 1947).

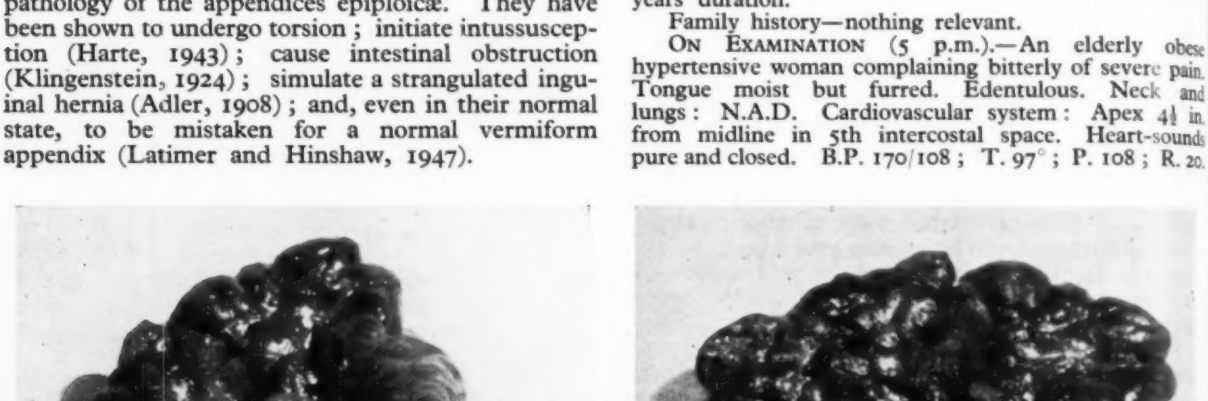

Fig. 356.—Twisted appendix epiploica. Note torsion at base and congestion of vessels.

Intra-abdominal torsion of an appendix epiploica is sufficiently rare to warrant the report of a further case. Attention was first drawn to the condition in a review by Hunt (1919), although the first clinical case was described by Riedel in 1905. Other reviews have been presented by Mabrey (1940); Pines, Rabinovitch, and Biller (1941); Giffin, McManamy, and Waugh (1942); and recently by Maranis and Cheek (1949) and Wakely and Childs (1949).

Two cases of interest are reported.

CASE REPORTS

Case 1.—

History.—A widow (M. P.), aged 72, was referred to hospital Feb. 25, 1952. She complained of a severe sharp abdominal pain of 54 hours' duration which radiated from the right lateral quadrant to the umbilical region and doubled her up. The onset had been sudden and there was nausea but no vomiting. On admission her pain was continuous and to the right of the umbilicus. Bowels were regular, with the daily use of aperients, but no blood or mucus had been passed. There were no genito-urinary symptoms.

Her previous history revealed that she was hypertensive; a uterine prolapse had been repaired some years previously and she had a flatulent dyspepsia of many years' duration.

Family history—nothing relevant.

On Examination (5 p.m.).—An elderly obese hypertensive woman complaining bitterly of severe pain. Tongue moist but furred. Edentulous. Neck and lungs: N.A.D. Cardiovascular system: Apex 4½ in. from midline in 5th intercostal space. Heart-sounds pure and closed. B.P. 170/108; T. 97°; P. 108; R. 20.

Abdomen: Tender with slight muscle guarding in an area lateral to the umbilicus but above the R.I.F. Bowel sounds present. P.R. and P.V. demonstrated no abnormality.

Urinalysis normal. Serum-bilirubin 0·2 mg. per cent. (Normal 0·1-0·8 mg. per cent.) Blood-count: Hb. 80 per cent; W.B.Cs 16,400; polymorphs 78 per cent; lymphocytes 20 per cent; monocytes 2 per cent.

A diagnosis of a doubtful cholecystitis with gall-bladder colic was made and she was put under routine observation. Two hours later on re-examination tenderness and guarding were noted in the right hypochondrium in addition to the right lateral quadrant. Beneath the guarding muscles a mass was thought to be felt and the above diagnosis was assumed to be correct. Seven hours after admission it was reported that the patient had had an attack of severe pain and the pulse-rate had risen to 120. Examination then showed that the tenderness had moved to the R.I.F. and across to the L.I.F. Rovsing's sign was positive. Hyperæsthesia was present in the R.I.F. and rectal examination revealed tenderness in the pelvis. The diagnosis was reviewed and a high-lying acute appendicitis was thought to have been missed, but the possibility of rupture of the gall-bladder was entertained.

AT OPERATION.—This was carried out forthwith under thiopentone, gas, and oxygen supplemented by pethidine and flaxedil (Dr. E. H. Franks). Through a small gridiron incision free serous fluid was discovered and a normal subcæcal appendix was found and removed. On further exploration there was revealed a large congested purplish appendix epiploica attached to the ascending colon by a narrow stalk which had undergone torsion. The pedicle had twisted some four times in an anti-clockwise direction. The twist was partially undone; the base transfixed and ligated and the mass removed. The wound was closed without drainage.

PROGRESS.—Convalescence was uneventful, although penicillin was given prophylactically in view of her obesity and the consequent danger of chest complications. Sutures were removed on the seventh day and the patient was discharged the next day.

PATHOLOGICAL REPORT (Dr. J. C. Valentine).—

1. Appendix 8 cm. long. Normal histologically and to the naked eye.

2. A roughly triangular piece of fatty tissue 4·5 × 0·5 × 0·7 cm. At one angle it had evidently become distorted, there being one and a half turns upon the pedicle (*Fig.* 356).

Histology: There is congestion and superficial infiltration of the area immediately beneath the peritoneum by polymorphonuclear leucocytes. The organ is composed entirely of adipose tissue with a slight fibrous tissue framework.

Case 2.—**Appendix Epiploica—Acute Torsion.** H. A. R., a male aged 68, was admitted on July 8, 1952, as an emergency with a volvulus of the sigmoid colon which was confirmed clinically and radiologically. An emergency Paul Mikulicz resection of the redundant loop was performed by Mr. D. W. Lacey on the same day. At the operation a small loose body was found in the pelvis. The patient made a good recovery immediately after the operation, but died on July 31, from an aspiration bronchopneumonia due to an inoperable carcinoma of the tongue.

At post-mortem examination (Dr. J. C. Valentine) no further loose bodies were discovered, and the origin from the colon could not be ascertained.

HISTOLOGY OF THE LOOSE BODY (*Fig.* 357).—A spherical nodule 0·8 cm. across, stony hard, and white in colour. On section it contains amorphous brown material and histologically the nodule is surrounded by a fibrous-tissue capsule, contained within which is a structureless material arranged around small spaces, giving a distinct suggestion of adipose tissue. I think the suggestion that this is a detached appendix epiploica which has calcified is correct.

DISCUSSION

Mechanism.—The cause of torsion has remained a mystery, but perhaps a combination of the many mechanisms suggested in the literature may be responsible. Pines and others (1941) think that obesity is one of the main predisposing factors in that the epiploic appendages are largest in the obese and in those who have recently lost weight. However, on reviewing 106 cases, obesity has been specifically noted in only 16. Morestin (1912) emphasizes that unusual length and size of the appendices were essential for torsion. A long or narrow pedicle with a bulbous free end is incriminated by Davis (1939) and this is noted here. Kantor and Hutcherson (1950) describe one 15 cm. long which simulated a twisted ovarian cyst. Black (1923) first suggested that the appendix epiploica may be the site of lipoma formation. This being the case, a lipoma at the free end would satisfy the condition of an expanded free end which is the essential factor. Although Pines notes that one of his specimens was short and stumpy and Taylor (1931) one 26 mm. long, this factor must be regarded as the most important local predisposing cause. Sudden exertion, trauma, and violent peristalsis have been blamed with no adequate proof. In fact only 4 cases

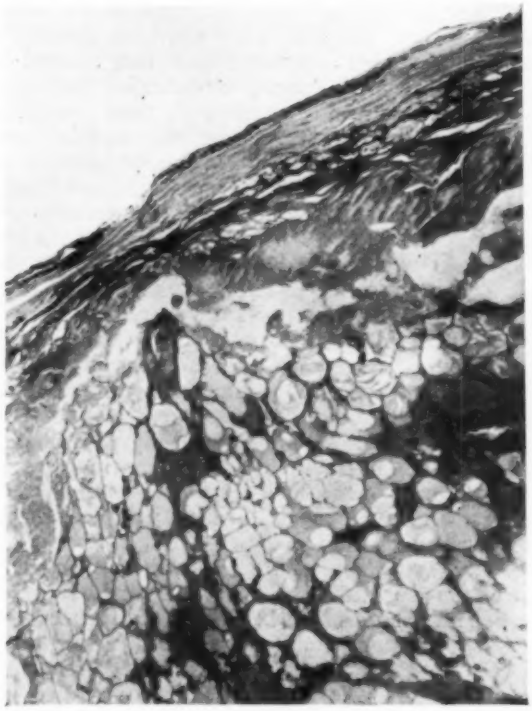

FIG. 357.—Photomicrograph of loose body, showing fibrous capsule containing accumulation of fatty cells. (× 60.)

have followed previous strain—after coughing (Fiske, 1936), after lifting (Colt, 1932), after delivery (Jenkins, 1949), and after currettage of uterus (Quillam, 1949). It has been noticed once during pregnancy (Walser, 1939). However in *Case* 1 it is observed that the patient was addicted to violent purgation. This fact may be a 'trigger' cause or perhaps merely a coincidence. The normal peristalsis is blamed by Wakely and Childs (1949).

Klingenstein (1924) holds the view that the primary cause develops from a distortion of a normal physiological function in that the twist is initiated by movement of an appendix towards a site of infection. However, such a source has been described in only very few reports.

Once the twist has begun the movement is kept up by 'hæmodynamic torsion', as postulated by Payr (1902). He described a plethora of the intestinal vascular system, normally present during the intestinal phase of digestion. This leads to a congestion of the veins of the appendix, which are longer than its central artery and which wind round it in a spiral fashion (Pines and others, 1941). The congestion imparts a torsion strain to the appendix and perpetuates the twisting movement.

This mechanism in the presence of a trigger factor, e.g., trauma or excessive peristalsis, and a predisposing factor, e.g., long pedicle and bulbous

free end, might well lead to torsion. However, it is remarkable, not that torsion occurs, but that it should not happen more often considering the state of continual movement within the abdomen.

Pathology.—The site of torsion is not always demonstrable, but when it occurs the result depends on the degree and rapidity of onset. There are six clear types which may be found in the literature.

1. *Acute Torsion.*—This is the result of a sudden acute episode. As the veins are compressed, the appendix becomes congested and œdematous, but with the onset of arterial obstruction an aseptic gangrene occurs. The appendix is thick, congested, cyanotic, and œdematous. In the extreme case, which is common in the literature, it is black and necrotic. Quillam (1949) reports a case where gangrene was advanced, although operation was performed within two hours of the onset of the pain. An obvious twist is seen at the base, varying from 180° (Zoeppritz, 1908) to ten and a half turns (Adler, 1908). The necrosed appendix may adhere to bowel and cause intestinal obstruction (Riedel, 1905), or may be cast off and adhere to nearby viscera (Mabrey, 1940). The peritoneal cavity contains serous fluid in the early stages, but it soon becomes hæmorrhagic with the onset of gangrene. Microscopically it is the site of intense congestion and œdema. Necrosis may be marked, and there is often an infiltration with acute inflammatory cells.

2. *Acute Torsion with Secondary Infection.*—A gangrenous appendix unless dealt with surgically may become secondarily infected, leading to a local abscess or a generalized peritonitis (Seelye, 1933). Harrigan (1917) quotes a case of Riedel from which a culture of *B. coli* was grown. The infection may originate by direct invasion from the bowel (Hunt, 1919) or from a diverticulum in relation to the base (Wakeley and Childs, 1949). The frequency of diverticula at the base of appendices epiploicæ is a point which is frequently overlooked, but was noted by Hamilton Drummond (1917) and Bland-Sutton (1903).

3. *Subacute Torsion with Adhesions.*—Torsion may occur to a minor degree with œdema and hæmorrhages in the organ, which may then adhere to a loop of bowel, its mesentery or another appendix forming a potential source of intestinal obstruction. Cases are reported by Klingenstein (1924), McIver (1933), Patterson (1933), and Moore (1940). Baumeister, Hargens, and Morsman (1938) describe a case of obstruction where three obstructing bands were present, the appendices at three separate sites being adherent to their fellow across the colon. Beattie (1950) notes two cystic appendices (*see below*) united by a strong fibrous band which could easily have caused obstruction.

4. *Chronic Torsion.*—The appendix may be the site of a chronic torsion. Le Vay (1941) quotes a case such as this with a 15 months' history of abdominal pain. Palmer and Hardman (1931) describe a case in which a cyst 5 cm. in diameter was formed in an appendix epiploica due to a slow torsion and a similar one was recorded by Hunt. Beattie's case is noted above. This type of torsion results in a fat necrosis which stimulates a chronic inflammatory reaction. Microscopy reveals areas of old hæmorrhage, fibrosis, and infiltration with chronic inflammatory cells.

5. *Loose Body Formation.*—Loose bodies, as distinct from foreign bodies, are on occasions found in the peritoneal cavity. Their origin has been disputed in the past and proves that there is no single source. Johnstone (1928) reports a hard calcareous mass lying free in the pelvis which, on microscopy, showed cubical epithelium resembling ovarian germinal epithelium. Sir James Simpson is reported to have found at post-mortem examination loose bodies, the origin of which were subperitoneal fibroids. Ovarian cysts are known to lie free in the pelvis. Ross and McQueen (1948) report a loose body which they suggest was foreign or inflammatory material from a perforated duodenal ulcer in which calcification had occurred. Most cases, however, are described as having their origin in the appendix epiploica. Deville (1851) reported the finding at an autopsy of five loose bodies and an appendix epiploica about to become detached from the transverse colon. However, he failed to associate the two conditions and it remained to Virchow (1863) to demonstrate the relationship between loose bodies and the appendices epiploicæ. He showed that the fat underwent saponification, with resultant calcification, and this process has been further investigated recently by Farr (1923). The increase in weight of the appendix caused strain on its pedicle, with obliteration of its blood-vessels. When this was complete, necrosis occurred and the appendix fell free into the peritoneal cavity. Perhaps torsion of the pedicle, especially of the chronic type, accomplishes the same end. Calcified loose bodies such as these are not common in the literature, but are reported by Littré (1703) and Riedel (1905), and by McIntosh (1947). Harrigan (1917) agreed that necrosis of the pedicle was the cause and quoted a loose body of degenerating fat, probably a twisted appendix which had become detached. Similar cases are reported by Pines and others (1941) and Mabrey (1940), and Hunt (1919) described a sac of degenerating fat free in the abdomen. On one hand the loose body was calcified, round, and smooth, and on the other it was fatty and necrotic, obviously the result of torsion. In the former case the histology is that of a central core of amorphous material surrounded by a fibrous sheath, but there is no direct histological evidence that it originated in an appendix epiploica or in fatty tissue.

Case 2 is of interest in this light in that fat cells are clearly seen in the histological section.

6. *Necrosis in Absence of Torsion.*—In some cases a necrotic appendix epiploica is found in which the base has no sign of a twist (Randall, 1932; Campbell, 1937; and Pines and others, 1941). Embolism or thrombosis with or without previous torsion is held responsible. Paul (1942) believes that in these cases traction from the weight of a fatty appendix causes tension and narrowing of the pedicle, with damage to the central vessels resulting in thrombosis.

Site of Torsion.—Necrosis may occur in the abdomen proper or in a hernial sac as a result of torsion (Riedel, 1905; Adler, 1908; Linkenfeld, 1908) or by strangulation at the constriction ring (Fitzwilliams, 1922; Ross, 1950). However, this

review concerns itself with intra-abdominal torsion. Most cases reported had a single appendix affected, but multiple lesions were described by Pines and others (1941), Hunt (1919), and Colt (1932).

The commonest site is the sigmoid colon, the incidence being almost double that of the cæcum. The rarest positions are the hepatic flexure and the descending colon (*Fig.* 358). On two occasions (Colt, 1932; and Virchow, 1863) the vermiform appendix was described furnished with appendices epiploicæ, and in Colt's case the appendix epiploica had undergone acute torsion with resulting gangrene. The site is important in the differential diagnosis as it explains the numerous and varied physical signs and symptoms.

Clinical Features.—Including the case reported here, 106 have been described and they have been reviewed. The incidence is almost equal in both sexes, there being 57 males and 49 females. No age was quoted in 5 cases, but the youngest was aged 18 and the oldest 76. The average age was 39·8 years and the maximum incidence between 30 and 50 (*Fig.* 359). The cases followed are divided into the following groups by a consideration of the symptomatology, duration of symptoms, and resulting pathology :—

1. Acute :—
   Primary 76.
   Recurrent 17.
   Acute on Chronic 4.
2. Chronic 6.
3. Incidental findings.

Without exception, the acute attack begins suddenly with severe abdominal pain, sometimes colicky, sometimes sharp. Although the pain is often experienced in the umbilical region, it varies usually with the site of the affected appendix. It may be in the left iliac fossa simulating a sigmoid diverticulitis or even a left-sided appendicitis

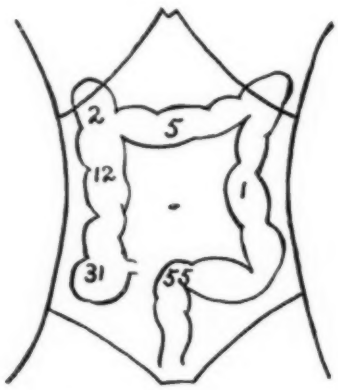

FIG. 358.—Sites of 106 cases from the literature.

(Byberg and Lam, 1945). It may be felt in the R.I.F. or in the right hypochondrium, but it is always noticed in an area corresponding to the contour of the colon. A shift of pain from the umbilical region to the site of the appendix was recorded in only 5 cases. Nausea and vomiting are unusual. Nausea was experienced in only 10 cases; vomiting in 13 and both in 11. If the lesion lies in the pelvis, frequency of micturition, tenesmus, and diarrhœa may be present. Rectal pain was noted once. However, the condition may be symptomless, as in 3 cases the torsion was discovered in the course of an operation for an unrelated and definite disease. Moore (1940) suggests that a minor degree of torsion may explain cases of mild abdominal pain which do not come to operation and for which no satisfactory explanation can be given.

The patient may report in the first attack but recurrent acute phases were noted in 17 cases. An

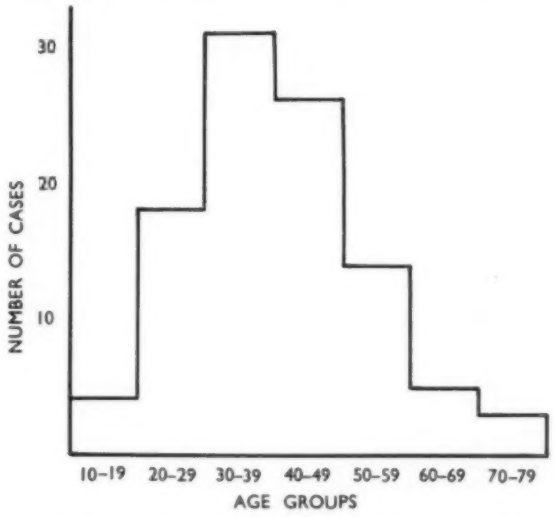

FIG. 359.—Age incidence (101 cases, 5 no age recorded; includes case reported aged 72).

acute flare-up may punctuate the sequence of an otherwise chronic illness. Most acute cases were referred to hospital late, the average case being 3·3 days old before admission was sought. Only 22 cases were admitted on the first day and a few on the twenty-first day. While it has been stated to occur in the obese and middle aged (Briggs, 1908), it was discovered that the commonest age period was 30 to 50 and that obesity was noted in only 16.

The patient does not look unusually ill. Tenderness over the affected site is invariable and this is common in the R.I.F., gall-bladder region, L.I.F. and on rectal and vaginal examination, depending on its situation. Guarding is present in only one-fifth of cases. A tender mass was palpated in only 20 per cent and its site and that of the tenderness has led to the multiplicity of conditions found in the differential diagnosis. It may be felt on rectal and vaginal examination and then may simulate pelvic disease. The pulse and temperature are elevated, but in the majority of cases where this was present peritonitis had developed. A leucocytosis was present in 23 out of 32 cases where the investigation had been carried out. This would suggest that secondary infection had occurred, but it was noted in some at an early stage (Maranis and Cheek, 1949).

In patients presenting a chronic condition, pain of a dull and constant character is the cardinal symptom. It is rarely severe and is referred to an area overlying the affected appendix. This may be suffered for as long as fifteen years before a medical attendant is consulted. Nausea and vomiting are absent. It is interesting to note that a mass can be

palpated in 66 per cent of these cases. Tenderness is constant, but there is no guarding or rigidity. The patient experiences little in the way of systemic upset.

Diagnosis.—The condition has been successfully diagnosed on only 2 occasions (Babcock, reported by Fiske, 1936; and Wakeley and Childs, 1949) the explanation being that the condition is rarely in mind and that there is no 'diagnostic train of symptoms' (Pines and others, 1941). It is rarely possible except in four sets of circumstances :—

1. When "the modest findings are paradoxical as related to the degree of subjective complaint" (Schimberg and Allen, 1947). It is frequently noted that the patient, who normally bears pain well, complains bitterly and yet the signs are remarkably slight. Note may also be made of the infrequency of nausea and vomiting.

2. When a mass is found in relation to the colon, with no preceding history to suggest diverticulitis, carcinoma of colon, cholecystitis, or ovarian disease. This state of affairs is liable to occur most often in chronic cases where time can be spent in investigation to exclude these conditions. Such a state of affairs was quoted by Le Vay (1941).

3. When a case presents with symptoms and signs suggestive of appendicitis, the appendix having previously been removed. This has occurred on three occasions (Hunt, 1919; Wakeley and Childs, 1949; and Kantor and Hutcherson, 1950).

4. When there is disproportion between abdominal tenderness, rigidity, and duration of symptoms (Pines and others, 1941). It is noted that there is marked tenderness with little or no guarding where from the duration of the disease marked guarding would be expected. This would seem to aid in excluding appendicitis.

On reviewing the pre-operative diagnosis it is found that almost all abdominal emergencies have been incriminated (*Table I*). This serves to emphasize the difficulty in diagnosis. Apart from the circumstances stated above, there are no clear-cut factors which will determine the diagnosis pre-operatively. Any diagnosis must be tentative.

Table I.—PRE-OPERATIVE DIAGNOSIS

	Cases
Acute or recurrent appendicitis	55
Laparotomy abdominal pain	13
Torsion ovarian cyst	6
Diverticulitis	7
Intestinal obstruction	4
Incidental findings	3
Carcinoma colon	3
Peritonitis	2
Cholecystitis	2
Correctly diagnosed	2
Tubo-ovarian abscess	1
Degenerating fibroid	1
Pelvic inflammation	1
Meckel's diverticulitis	1
Acute epiploitis	1
Abdominal colic	1
Abdominal wall abscess	1
Acute left-sided appendicitis	1
Torsion of intra-abdominal testis	1

Treatment.—This is simple but imperative in order to prevent secondary infection and its sequelæ. The twisted base is transfixed and ligated and the appendix removed. Care is taken to apply the suture a little way from the colon as a diverticulum may be present in relation to the base of the appendix and may be damaged. For such a reason it has been recommended that the raw area be reperitonealized by a purse-string suture. (Rankin, Bargen, and Buie, 1932.) In addition the operator may ligate the loop of the vasa recta which projects into the base of the epiploic appendage and from which its central artery arises, with subsequent damage to the wall of the colon. Drainage is not usually necessary provided that there is no local abscess.

Prognosis.—The outlook is good. In fact Mabrey (1940) records a case where the twisted appendix was left in situ at operation and complete recovery ensued. However, 4 fatalities were noted before the syndrome was much publicized. The causes of death were peritonitis in 2 cases, intestinal obstruction in 1 case, and paralytic ileus due to a "secondary coagulative process in the mesenteric vessels" in 1 case. (Riedel, 1905; Ebner, 1909, Klingenstein, 1924; and Seelye, 1933.) Early operation has been a feature of cases reported in the past twenty years and has been a factor in preventing further fatalities.

SUMMARY

1. Two cases are described, one of intra-abdominal torsion of an appendix epiploica and one of intraperitoneal loose body originating from an appendix epiploica.

2. A review of 106 traced cases of intra-abdominal torsion of an appendix epiploica is reported.

3. The mechanism of acute torsion is briefly discussed.

4. A classification of pathological varieties and clinical types has been suggested.

5. An attempt has been made to clarify the circumstances under which a correct diagnosis can be made.

My thanks are due to Mr. G. J. Griffiths for permission to treat the first case; Mr. D. W. Lacey for the details of the second case; Dr. J. C. Valentine for the pathological reports; Mr. Jewell Harrison for the photographs; and the Department of Pathology, University of Bristol, for the photomicrograph.

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KAOLIN GRANULOMA

BY R. ERIC POTTS

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WHILE the occurrence in skin and subcutaneous tissue of granulomata caused by the implantation or impaction of silicious particles is recognized, it has not received the prominence accorded to the analogous lesions of intra-abdominal tissues. In contradistinction to the latter, however, the silicious granulomata of skin and subjacent tissues are not confined to those produced by talcum powder. Thus, Shattock (Shattock and Dudgeon) (1916) reported a local silicious granuloma due to impacted stone many years before talcum-powder granuloma was first recognized by Antopol (1933). It is the purpose of this paper to call attention to the occurrence of a further variety of local silicious foreign-body reaction, namely, kaolin granuloma.

CASE REPORTS

Case 1.—

CLINICAL SUMMARY.—C. W., a nurse aged 22 years, complained of pain in and discharge from the left breast. She stated that sixteen months previously she had developed a painful swelling above and medial to the nipple and that following rupture of this inflammatory mass, a sinus had formed. Kaolin poultices were applied on several occasions during this time but the lesion failed to heal, although temporary improvement occurred during penicillin therapy. A wedge of tissue including the sinus was excised on Feb. 7, 1952. There had been no previous operative treatment and no dusting powder had ever been applied to the area. The patient gave an excellent history.

PATHOLOGY.—The specimen comprised a piece of fibro-fatty tissue 27 × 24 × 16 mm. covered by skin.

Histologically, the subcutaneous tissue was largely replaced by a chronic inflammatory mass. Superficially, this granulomatous lesion extended up to, and at one area ulcerated the covering epithelium, while laterally and deeply it merged into the dense collagenous scarring of the surrounding tissue. In the granulation tissue the major part of the cellular infiltrate was formed by lymphocytes, plasma cells, and large mononuclears, but polymorphonuclears were present at some areas. The most striking feature of the tissue reaction was, however, the presence of moderately-sized multinucleate giant cells in which the irregularly-distributed nuclei seldom exceeded ten in number (*Fig.* 360). These cells, smaller in size but otherwise similar to the foreign-body type of cell commonly encountered, showed small translucent cleft-like spaces in the cytoplasm when examined by direct illumination: crossed polaroid screens revealed these as birefringent crystalline particles. Using these screens it was also apparent that there was a significant correlation between the occurrence of the giant cells and the distribution of the doubly refractile material. The latter was not, however, confined to the multinucleate cells, since a few of the large mononuclears also contained similar particles, but almost all the crystals were intracellularly situated in one or other of these two cell types (*Fig.* 361). The crystals themselves were uncoloured and most frequently of spicular contour, although their small size, which was of the order of 10 microns, precluded a detailed study of their form. All resisted immersion in *N* hydrochloric acid for 30 minutes, and exposure to 600° C. for 10 minutes.

Case 2.—

CLINICAL SUMMARY.—M. M., a 30-year-old housewife, complained of a recurrence of inflammation in the coccygeal region. A pilonidal sinus had been discovered in 1942. She had suffered the usual recurrent inflammatory episodes and had applied kaolin poultices in 1949. The affected area was excised on March 30, 1952.

PATHOLOGY.—The specimen comprised a skin-covered portion of tissue 45 × 20 mm. in surface extent and 15 mm. in depth.

Histologically, this showed a small sinus extending down into the subcutaneous tissue, lined in part by squamous epithelium and in part by chronic non-specific granulation tissue. The dermis and subcutaneous tissue adjacent to the sinus were widely involved by fibrous

scarring, in the mature collagenous tissue of which were scattered cellular foci consisting of lymphocytes, plasma cells, and large mononuclears, together with a variable but generally small number of giant cells. These latter were uniformly of foreign-body type and frequently contained tiny spicular crystals inconspicuous by ordinary illumination, but brightly visualized between crossed polaroid screens (*Fig.* 362). All these crystalline inclusions resisted exposure to *N* hydrochloric acid for 30 minutes and exposure to 600° C. for 10 minutes.

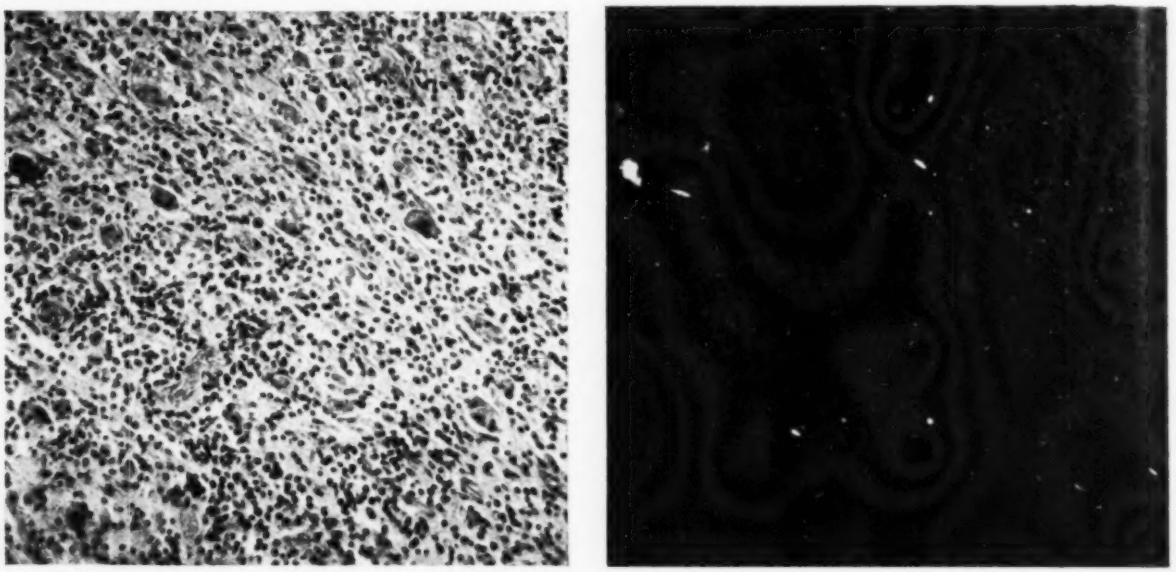

FIG. 360.—A representative field of the lesion in *Case* 1 by ordinary illumination and through partially crossed polaroid screens. In the chronic granulomatous tissue reaction several small multinucleate giant cells are present, some of which are seen to contain birefringent kaolin crystals. Many particles are, however, outwith these giant cells. Hæmatoxylin and eosin. (× 140.)

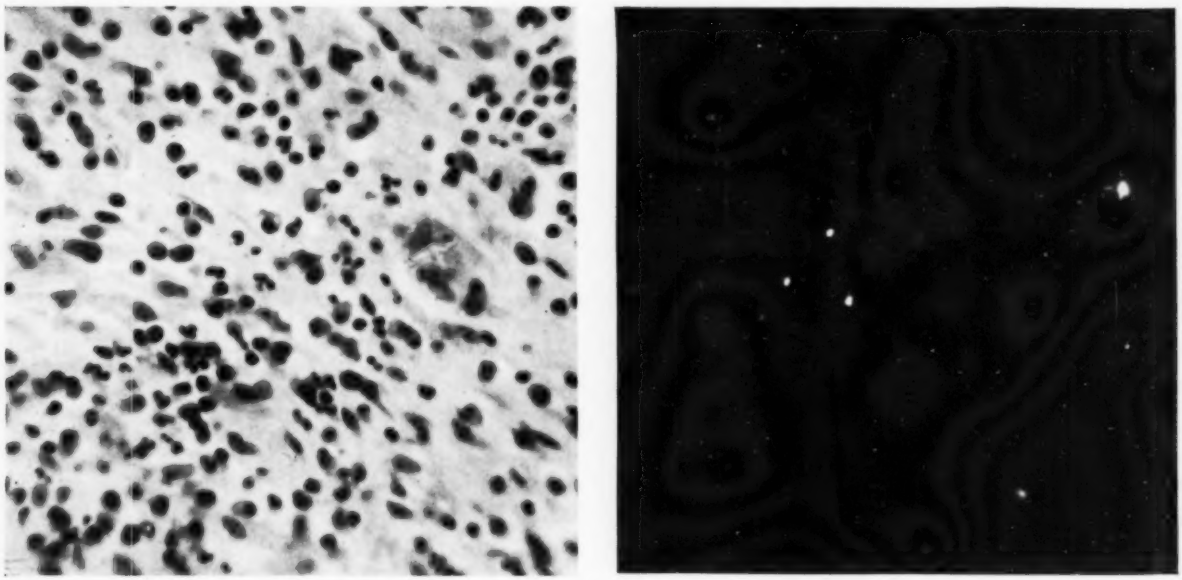

FIG. 361.—A higher magnification of the lesion in *Case* 1 by ordinary illumination and through partially crossed polaroid screens. This shows the intracytoplasmic situation of the majority of the crystals within giant cells or large mononuclears. A number of polymorphonuclear leucocytes are present in this field. Hæmatoxylin and eosin. (× 400.)

DISCUSSION

Commentary on the Cases.—In view of the frequency with which kaolin poultices are used in the treatment of inflammatory diseases of skin and subcutaneous tissue, it is remarkable that the occurrence of local foreign-body granuloma due to this substance has not, so far as I am aware, been reported hitherto. It does not, however, follow that the development of silicious granuloma in such cases is a rarity, for only those lesions which fail to heal after a very long time reach the surgeon, while still fewer are subjected to excision or biopsy and ultimately pass to the pathologist. Moreover, kaolin crystals are inconspicuous in the tissues and are readily overlooked unless the section is examined between crossed polaroid screens, and even if

recognized they are likely to be regarded as talc. The two varieties of crystal are, indeed, very similar in that they are both spicular in form and of the order of only a few microns (*Figs.* 363, 364). Kaolin crystals are somewhat smaller than those of talcum powder but the difference is not sufficient to provide a reliable morphological distinction. Both are colourless by direct illumination and doubly refractile between crossed Nicol's prisms, but it should be mentioned that by no means all the particles in either talcum powder or kaolin powder are birefringent, even when examined at various degrees of rotation on a rotating stage.

It is not surprising that the two types of crystal elicit a similar tissue reaction since both are naturally-occurring insoluble silicates. Talcum powder consists largely of hydrated magnesium silicate ($3MgO.4SiO_2.H_2O$) while kaolin is mostly composed of hydrated aluminium silicate ($Al_2O_3.2SiO_2.2H_2O$).

The cellular reaction in the two cases described is closely similar to that regularly encountered in granulomata due to talcum glove powder, and both were, in fact, initially regarded as instances of talc granuloma until subsequent inquiries excluded the possibility of ingress of such material into the tissues and established at the same time a clear history of repeated applications of kaolin.

The role of the implanted kaolin as a factor in the failure of healing in the second of the two cases is speculative, since chronicity of the inflammation in

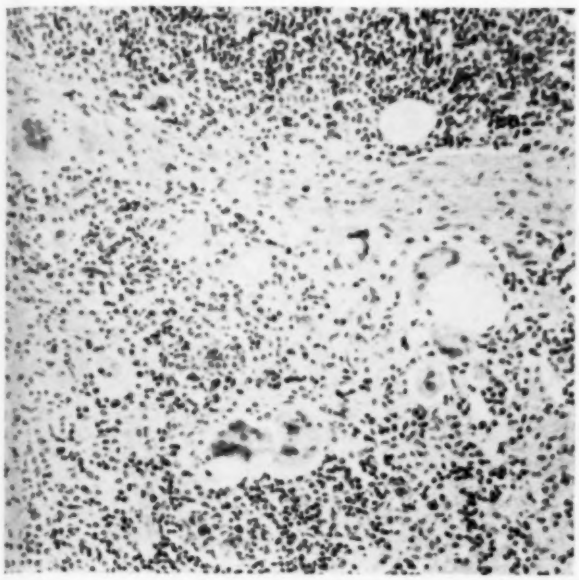

FIG. 362.—The lesion in *Case* 2 photographed by ordinary illumination and through partially crossed polaroid screens. The giant cells are larger than in *Case* 1 and lymphocytes are more conspicuous. Doubly refractile crystals are again seen, both within the multinucleate giant cells and outwith them. Hæmatoxylin and eosin. (× 140.)

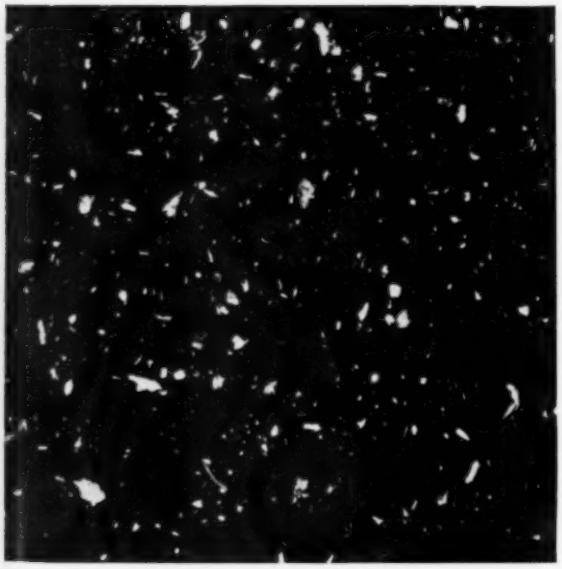

FIG. 363.—Kaolin powder photographed through crossed polaroid screens. (× 140.)

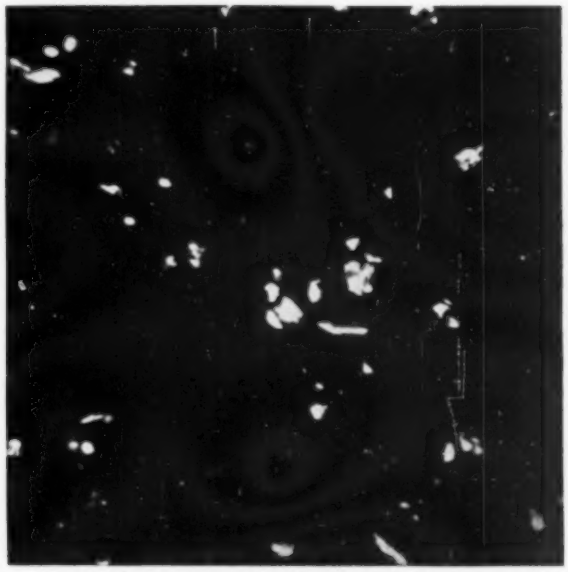

FIG. 364.—Talcum powder photographed through crossed polaroid screens. (× 140.)

pilonidal sinuses is the rule rather than the exception. In the first case, on the other hand, there seems little doubt that the presence of the kaolin crystals and the resultant foreign-body reaction was responsible for the maintenance of the sinus—a view which is consistent with the very temporary improvement during penicillin therapy.

It is known that kaolin gives rise in the tissues to a chronic inflammatory lesion followed by fibrous scarring, and it has been used for the purpose of obliterating the pleural cavity (Baader, 1950). It is also of interest that pneumoconiosis due to long exposure to kaolin dust has been described in two pharmaceutical workers, although the diagnosis was clinical (Tara and Trouard-Riolle, 1948). Nevertheless, I considered it desirable to confirm the granuloma-eliciting property of kaolin by inoculation into the tissues of the experimental animal. Examples of these artificially-produced lesions are shown in *Figs.* 365 and 366. They left no doubt of the capacity of kaolin to elicit a foreign-body giant-cell reaction: indeed, the early lesions consisted predominantly of multinucleate giant cells with an admixture of large mononuclears, while, in contradistinction to

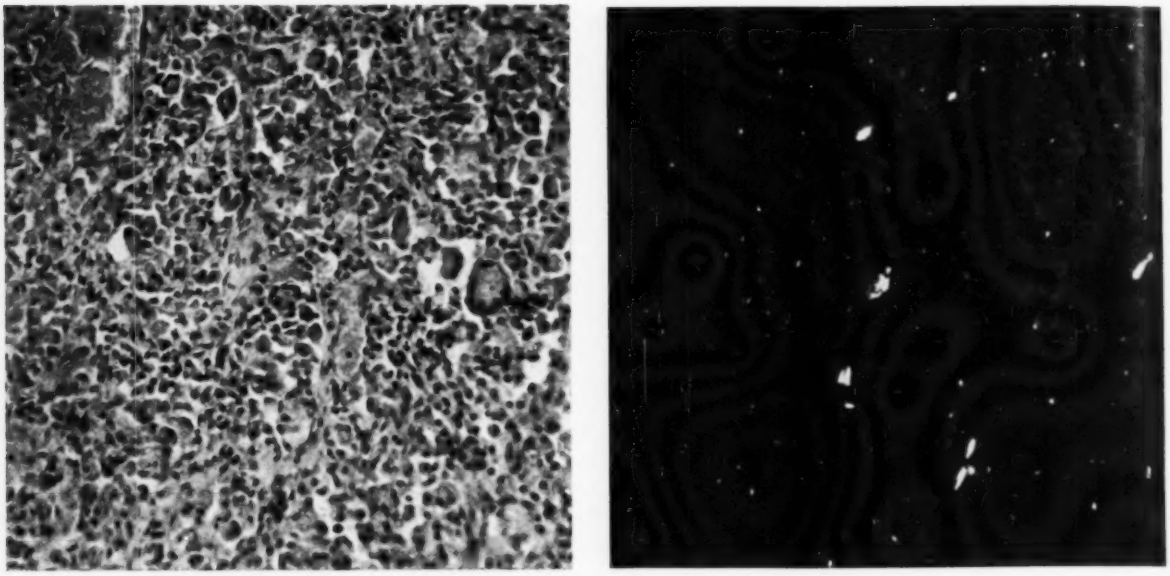

FIG. 365.—Experimental lesion in the guinea-pig, three weeks subsequent to subcutaneous injection of kaolin in aqueous suspension, by ordinary illumination and through partially crossed polaroid screens. The lesion consists almost entirely of binuclear and small multinuclear cells with a smaller proportion of mononuclear histiocytes. The Langhans-like cell on the right-hand side was one of many in this tissue. The crystals are inconspicuous by direct illumination, but are prominent between polaroids. The majority are clearly intracellular and in most instances very small, although several larger particles are also seen. Hæmatoxylin and eosin. (× 140.)

FIG. 366.—Experimental lesion in the guinea-pig seven weeks after subcutaneous injection of aqueous suspension of kaolin, by ordinary illumination and through partially crossed polaroid screens. This shows a similar histological picture to the three-week lesion with the addition of early fibrosis. Iron hæmatoxylin-picro-acid fuchsin. (× 140.)

the human cases, lymphocytes and plasma cells were present in negligible numbers and no granulocytes were found. The experimental granulomata were, however, closed lesions, free from the effects of secondary pyogenic inflammation which existed in the human cases described. There was no significant fibrosis in the earlier lesions, but at seven weeks there was appreciable fibroblastic and, at places, more collagenous scarring. In short, there remained no doubt that kaolin might be responsible for a silicious granuloma very similar to that produced by talc.

Differentiation from Talc Granuloma.—Although in a lesion established as a silicious granuloma the differentiation of kaolin and talc as the aetiological agent may be obtained by careful inquiry, it would be advantageous to have available a simple method for the independent identification of the two types of crystal in tissue sections. Both are highly insoluble substances and resist immersion in mineral acids in all strengths, but at the same time insolubility in the strictly chemical sense is commonly relative rather than absolute, and such is the case with hydrated aluminium silicate. It seemed, therefore, feasible that a sufficient number of aluminium or silicate ions might be available in solution to visualize one or other of these radicles. Alizarin sulphonate is a sensitive although not specific reagent for the detection of aluminium, while the acidified ammonium molybdate-benzidine technique is specific for silicates in the absence of phosphates and with the dismissal of arsenates and germanates. Initial 'spot-test' trials with kaolin powder on filter-paper failed to show any perceptible colour with alizarin Red S in either aqueous or alcoholic solution and this test therefore was not further considered. On the other hand, with the acidified-molybdate method the formation of silico-molybdate from the kaolin was readily visualized by benzidine reduction, while no reaction was obtained with parallel tests using talc, so that the method clearly offered prospects of successful adaptation to the differentiation of the two substances in tissue sections. This expectation was not, however, fulfilled: microscopically there was no appreciable staining of the kaolin crystals in either the human or experimental lesions, despite the fact that in the sections of the more heavily kaolin-impregnated animal tissues a faint but unequivocal blue coloration was evident on naked-eye examination. It is, unfortunately, a serious shortcoming of the very highly sensitive molybdenum blue method that the coloured end-product is subject to considerable diffusion; while ammonium silico-molybdate, unlike the corresponding phospho-molybdate, is soluble in acid. Furthermore, a blue coloration is not readily detected microscopically in small amounts even if the illumination is reduced to take advantage of the Purkinje effect. For the moment, therefore, there appears to be no method, other than by direct inquiry, of identifying as kaolin small amounts of doubly refractile silicious material such as are found in the tissues.

Differentiation from Proliferative Tuberculosis and Sarcoidosis.—In the wider differential diagnosis of kaolin granuloma, it should be borne in mind that talc may give rise to a follicular granuloma bearing a histological resemblance to tuberculosis or sarcoidosis, especially where the quantity introduced into the tissues is small and widely scattered. This similarity is prominent, too, in some cases of traumatic silicious granuloma due to stone particles and, indeed, was responsible for the designation 'pseudotuberculoma silicoticum' applied to this type of lesion. It seems likely that kaolin might elicit a similar reaction, and attention is therefore drawn to the fact that intracellular non-silicious doubly refractile crystalline inclusions may occur in both sarcoidosis (Sklavlem and Ritterhoff, 1946) and hyperplastic tuberculosis (Potts, 1951). In this connexion it is of differential diagnostic importance that the endogenous crystals of both tuberculosis and sarcoidosis are always soluble in *N* hydrochloric acid within half an hour, in contradistinction to silicious crystals, whether kaolin or talc.

SUMMARY

Two cases of subcutaneous kaolin granuloma are described. The lesions are morphologically indistinguishable from those caused by talc. Similar lesions, produced by the injection of kaolin in experimental animals, are illustrated.

The chemical differentiation of kaolin and talc crystals was achieved in macroscopic spot tests, but the same method was not successful in tissue sections.

Kaolin, like talc, is resistant to dilute mineral acid, and in consequence is readily differentiated in the tissues from the endogenous birefringent crystalline inclusions of tuberculosis and sarcoidosis.

I am indebted to Dr. W. B. Davis for his helpful suggestions in the preparation of the script. Mr. R. Cormack and Mr. G. Ralston kindly arranged for detailed clinical histories. The photomicrographs are the work of Mr. H. Gray.

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THE RELATIVE SUSCEPTIBILITY TO INJURY OF THE MEDIAL AND LATERAL POPLITEAL DIVISIONS OF THE SCIATIC NERVE *

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It is the experience of most observers that in injuries of the sciatic nerve the lateral popliteal division is more frequently involved and more often suffers greater damage than the medial (Stookey, 1916; Benisty, 1918; Hammond, 1918; Medical Research Council Report, 1920; Stookey and Scarff, 1943). The object of this paper is to examine this observation in the light of recent work on the internal anatomy of peripheral nerve-trunks.

In a series of 67 injuries of the sciatic nerve and its popliteal divisions from the author's series there were 48 gunshot and stretch injuries which occurred at a level which permitted a determination of the relative vulnerability of the two divisions of the nerve to mechanical violence. Measurements of the severity of the lesion when both divisions sustained injury were based on the delay intervening between injury and the onset of recovery, the progress of recovery, and the nature of the residual disability. An examination of the clinical data in this way revealed the following information :—

Effects approximately the same in the fields of both nerves	16
Effects greater in the field of the medial popliteal division	6
Effects greater in the field of the lateral popliteal division	26

In some cases the situation of the entry and exit wounds indicated that the involvement of one or other division of the sciatic nerve could have been due to the track taken by the missile. It was quite clear, however, that the higher incidence of greater and selective damage to the lateral division could not be attributed to this factor. Thus these findings proved to be consistent with those reported by others.

What is the explanation of the greater vulnerability of the lateral popliteal division to damage when the sciatic nerve is injured ? Though this clinical observation has been the subject of repeated comment, a convincing explanation to account for the phenomenon has yet to be presented. The object of this paper is to examine this question with particular reference to recent work on the internal anatomy of peripheral nerve-trunks.

Relative Positions of the Two Divisions in the Sciatic Trunk.—Stookey and Scarff (1943) believed that the peroneal (lateral popliteal) division was much more likely to suffer than the tibial (medial popliteal) division in injuries of the sciatic trunk because the former is placed " more superficial than the tibial, owing to the fact that the sciatic is somewhat rotated ". This arrangement, however, obtains only in the buttock and upper fourth of the thigh and the extent to which the lateral division overlaps the medial posteriorly (i.e., superficially) is very limited (Sunderland and Ray, 1948). Though this anatomical feature could be a contributing factor in some injuries, it is unlikely to be solely responsible in the majority. Furthermore there were cases in the series investigated by the author in which the missile had passed closer to the medial division than to the lateral, with greater damage to the latter.

Blood-supply.—Hoffman (quoted by Hammond) believed the greater susceptibility of the lateral popliteal division to be due to a poorer blood-supply, but an investigation (Sunderland, 1945, a) of the vascular pattern of the sciatic nerve failed to reveal any features which were specific to the lateral division or which would render it more vulnerable to injury. In an investigation of the paralysis induced by direct pressure and by tourniquet in the sciatic nerve of the cat, Denny-Brown and Brenner (1944) observed that the lesions produced in their experiments were maximal in the peroneal division just below the bifurcation of the sciatic nerve. They compared this observation with the reported differences in vulnerability of the medial and the lateral popliteal nerve in man to apparently similar injurious agents and explained the relative susceptibility of the two branches as follows : " Besides the obvious difference in size of the two nerves the peroneal division is usually in the form of one major bundle, so that its vessels have less protection than those of the popliteal division, which lie in the crevices between the bundles. This anatomic difference may certainly account for the difference in lesions caused by pressure."

It is felt that though such a vascular factor may operate in some cases this is unlikely to be responsible in most cases in man, particularly in those lesions which are not caused by pressure. Although the fibres of the lateral popliteal division may be collected into a single funiculus, this is an unusual arrangement which, when present, does not involve the entire length of the nerve. The funicular pattern is usually such that the intraneural vessels do not appear to be more exposed to injury than they are in the neighbouring medial division. As we shall see, however, the funicular pattern of the nerve does present features which contribute to the greater vulnerability of the lateral popliteal division.

The Effect of the Size, Number, and Disposition of the Funiculi on the Consequences of Traumatic Nerve Injury.—It is uncommon for human peripheral nerves to be composed of a single funiculus. They are, on the contrary, usually composed of several funiculi which by their repeated division and anastomosis engage in plexus formations along the length of the nerve. As the result of these changes, the number, size, and arrangement of the funiculi vary from level to level. In addition to these variations, peripheral nerves present individual lateral and regional differences and peculiarities as regards the proportion of the cross-sectional area of the nerve-trunk occupied by its funiculi. The supporting epineurial tissue likewise varies in amount

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along the length of the nerve. This information has been detailed elsewhere (Sunderland and Bradley, 1949), and for the purposes of the present discussion it will suffice to state that the funiculi of human nerves are more compactly arranged in some regions and in some nerves than in others and that in some specimens the bundles are numerous but small and widely separated.

On the basis of variations in the relative amounts of funicular and connective tissue it is conceivable that nerves, or those segments of nerves which are composed of large and tightly packed funiculi, are more vulnerable to mechanical injury than those in which the bundles are smaller and are more widely separated by a greater amount of connective tissue. In the former the forces acting on the nerve are concentrated on the main content of the nerve, namely the funiculi. In the latter they are broken up and dispersed by the connective-tissue packing while the funiculi, being more loosely arranged, are more easily displaced within the nerve and this reduces the effects of deforming forces.

An additional advantage is enjoyed by the nerve which is composed of numerous small funiculi when it is subjected to mechanical trauma which falls unevenly across the nerve. Under these circumstances the tendency is for some funiculi to escape while others are involved in varying degree. In partial injuries of this nature the intraneural damage will be confined to the bundles lying within the field of the violence. The tendency, then, is for such lesions to be self-limiting. On the other hand, when the bundles are few in number and large in size the violence is concentrated on all or the majority of the fibres comprising the nerve, while the rupture of intrafunicular vessels (which may be very large in large bundles) and a fibroblastic reaction may lead to complications which can seriously embarrass large collections of fibres.

An investigation of the cross-sectional area of the sciatic trunk and its popliteal divisions devoted to funiculi and to connective-tissue packing has demonstrated that in most individuals the lateral popliteal is composed of fewer and larger bundles and less connective-tissue packing than is the medial division. This morphological difference, which is clearly illustrated in the accompanying graph (*Fig.* 367), must be a significant factor in rendering the lateral division more liable to injury. Furthermore it has also been shown that the lateral popliteal nerve usually contains less adipose tissue in the epineurium than does the medial division (Sunderland, 1945, b.)

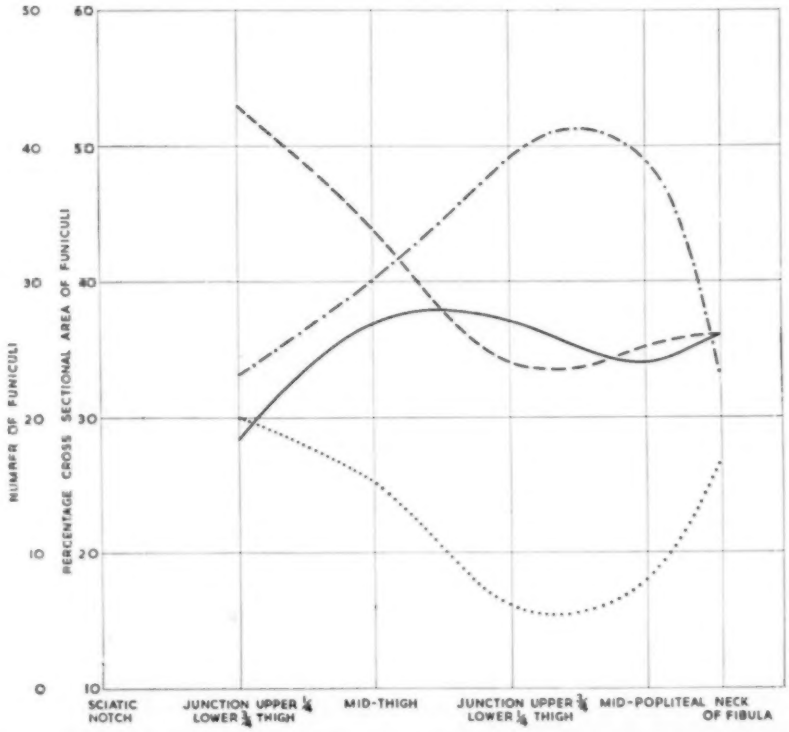

FIG. 367.—Graphic representation of the average percentage funicular area and the average number of funiculi based on measurements at five levels in twenty specimens of the sciatic nerve. Medial popliteal division: Percentage funicular area, ————; Number of funiculi, — — — . Lateral popliteal division: Percentage funicular area, —.—.—; Number of funiculi, · · · ·.

Course pursued by the Two Divisions between the Sciatic Notch and the Knee.—A further point of interest concerns the course taken by the two divisions in the thigh. The medial division descends more vertically from the sciatic notch and continues into the leg with little fixation as the result of the entry of branches into the muscles innervated by it. Consequently when this division is subjected to violent lateral displacement, such as is caused by the 'near miss' passage of a high-velocity missile, the nerve is not stretched between two fixed points, and the force is consequently dissipated over a greater length of the nerve. On the other hand, the lateral

popliteal division descends more obliquely in the thigh and is angulated and relatively securely fixed at both the sciatic notch and the neck of the fibula. Therefore when this nerve undergoes violent lateral displacement as the result of a 'near miss' by a high-velocity missile or explosive bullet, the sudden stretch to which the nerve is subjected involves a length of nerve which is strung between two relatively fixed points. There is consequently less slack to take up the displacement and the nerve is therefore subjected to greater stretch. This in turn means a more severe injury, particularly when the nerve contains a small amount of supporting connective tissue.

SUMMARY

1. The observation that in injuries of the sciatic nerve the lateral popliteal division is more frequently involved and more often suffers greater damage than the medial is confirmed.

2. It has been argued that the large size of the funiculi which are also fewer in number and the relatively smaller amount of supporting connective tissue in the lateral popliteal, as opposed to the reverse arrangement in the medial division, is the principal factor contributing to the greater susceptibility to injury of this division of the sciatic nerve.

3. An additional anatomical feature concerns the course taken by the two divisions in the thigh. The lateral division is fixed and angulated at the sciatic notch and neck of the fibula. When the nerve is subjected to violent lateral displacement it is stretched between these two unyielding points and in this way is damaged. The medial division is less securely fixed inferiorly where its relations are such that during lateral displacement the stretch can be dissipated over a greater length of the nerve.

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POLYORCHIDISM

A REPORT OF TWO CASES WITH TORSION

BY W. A. WILSON AND J. LITTLER, LIVERPOOL

POLYORCHIDISM has roused man's interest and imagination from the earliest times. At first it was supposed that it endowed him with supernatural fecundity, and in the literature of olden times there are many reports of men with four, five, and even six organs. However, as time went on, and more accurate observation has taken the place of old-time superstition, the number of cases has diminished, and interest now is focused more on the rarity of the condition.

HISTORICAL REVIEW

Although Blassius is reported by Marriotti (1907) as having found the condition in the course of a post-mortem examination in 1670, the generally accepted first true report of polyorchidism is that of Ahlfeld (1880). This, too, was a post-mortem finding, and to Lane (1895) falls the distinction of first reporting a case found at operation, and later proved histologically.

Polyorchidism was reported several times, and with varying degrees of accuracy, during the first quarter of this century, until in 1928 Eddington and Blacklock reviewed the condition. They collected all available reports, and analysed them, only accepting as true instances of the condition those cases in which the supernumerary organ had been proved a testis by histological examination. This standard has been universally accepted since. They instance only 8 cases so proved, and add their own. They do not mention Ahlfeld's case, but it has been accepted by subsequent authors, and we feel it should be included since it does fulfil the criterion of histological proof. Oudendaal's (1922) peculiar case is mentioned by Eddington and Blacklock. Here, although the mass was proved to be testicular on histological examination, it was found intra-abdominally, suspended from the ileum, about 30 cm. from the ileocæcal valve, and probably for this reason was not considered to be within the scope of their paper. It has not been mentioned by any subsequent author, and we feel, therefore, that we should not include it in our review.

King (1931) and Boggan (1933) each reported a case, and Boggan, accepting Ahlfeld's one and including his own, stated that 12 proven examples of polyorchidism had been reported to that date. Jordan and Dodson reported a further case in 1934, but the histology of the lower nodule showed only epididymis. Although their explanation of this (that the testicular tissue had atrophied following an injury some fifteen years previously) is ingenious, we feel that this might be a case of a detached part of the epididymis, and that it should not be accepted as a true instance of polyorchidism.

Since then Chevassu (1936), Goldstein and Cassilli (1939), Cranz (1942), Thiessen (1943), Handley and Crawford (1944), Dardill and Constantini (1947) (three cases but only one proved histologically), and Smith, Evans, and Suder (1950) have

all reported cases which have been proved by histological examination, and are, therefore, acceptable. There are, therefore, including Ahlfeld's, but excluding Oudendaal's and Jordan and Dodson's cases, 19 proven examples of the condition reported in the literature to date.

We wish to add 2 further examples which fulfil the conditions laid down by Eddington and Blacklock, making 21 cases in all.

CASE REPORTS

Case 1.—A. M., a labourer, aged 35, married with five children reported to the Out-patient Department of the Birkenhead General Hospital, complaining of a painful swelling in the left groin. The swelling had been present for ten years. It appeared after heavy work, and in the evening. Usually it had gone by morning. Occasionally, and more frequently during the few months prior to attendance at hospital, it had been painful for a day or two at a time. Previous medical history was of a posterior gastro-enterostomy for a chronic stenosing duodenal ulcer in 1943.

On Examination.—A healthy, well-built male. Well compensated mitral stenosis, with a mitral thrill, and a presystolic murmur. Sound, well-healed, upper midline scar. There was a left inguinal hernia with an impulse extending as far as the external ring. There was also a thickening of the spermatic cord in this region. The left testis and epididymis were small and high up in the scrotum. The right side was normal. He was recommended operative treatment for his left inguinal hernia, and was admitted to hospital.

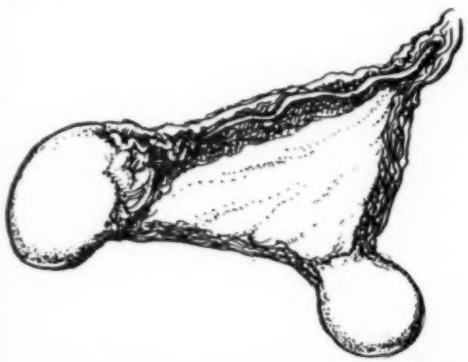

Fig. 368.—Diagram showing the supernumerary testis with the cords and triangular membrane as described in the text.

At Operation (March 2, 1949).—The left inguinal canal was laid open. There was an indirect inguinal hernial sac. This enveloped a mass, approximately 3½ cm. by 2½ cm., lying in the canal and apparently continuous with the spermatic cord. The sac was dissected off the cord and the mass, to which latter it was adherent; it was then transfixed, ligated, and excised.

The mass was now more closely examined. It appeared to be an extra testis and epididymis with a cord of its own which joined the cord of the normal testis at the level of the internal inguinal ring. There was a thin triangular sheet of tissue between the two testes and their cords. The cord to the lower testis was normal, with a vas, but only a thin cord containing vessels with no apparent vas ran to the upper one. In the base of the sheet of tissue joining the two testes there was also a thickening which seemed to consist of vessels only. (*Fig.* 368.) The cord to the upper testis was divided at the internal ring, and this testis was removed (*Fig.* 369). The lower testis was fixed to the bottom of the scrotum. The hernia was repaired by narrowing the internal ring, and darning the conjoint tendon to the recurved edge of the inguinal ligament with nylon. The external oblique aponeurosis was overlapped behind the cord. The patient had an uninterrupted convalescence, getting up on the third post-operative day, and he was discharged home on the twelfth day.

Microscopical Appearance (*Fig.* 370).—The specimen is testis. The seminiferous tubules show dilatation, are atypical in formation, and show no spermatozoa.

Fig. 369.—Operation specimen from *Case* 1.

There has been recent hæmorrhage into the testicle, but this has remained comparatively localized, and in this area endarteritis obliterans is seen. There is no evidence of malignancy or acid-fast disease. The cord shows a plexus of vessels and nerves. The vas deferens is not present.

Case 2.—E. S., aged 44, married, no children. The patient complained of fairly severe pain, and of swelling in the right groin, of twenty-four hours' duration. There had been a similar attack four years previously and a number of less severe episodes in the intervening period. Trauma had not been a precipitating factor at any time. There were no relevant previous illnesses.

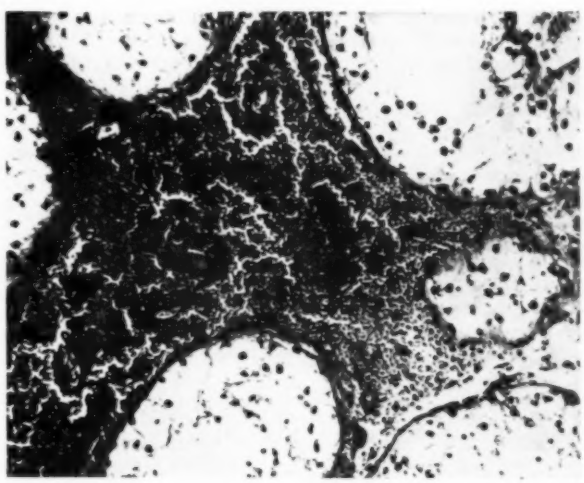

Fig. 370.—*Case* 1. Microscopical appearance. (× 186.)

On Examination.—The left testis and epididymis were normal in every way. The right side of the scrotum was empty. In the right groin there was a tender swelling apparently lying in the inguinal canal. There were no other abnormal findings, and it was decided that this swelling was an undescended right testis which had undergone recurrent minor attacks of torsion. The patient was admitted to Birkenhead General Hospital

and at operation on May 27, 1950, a right inguinal incision was made, and the external oblique aponeurosis incised and reflected. Just within the external abdominal ring lay the testicle. In its spermatic cord was an apparently normal vas deferens. There was a large associated indirect hernial sac. The sac was separated from the cord, and traction put upon the latter, whereupon another

FIG. 371.—Operation specimen from *Case* 2. (× ½.)

well-formed testis and epididymis appeared through the internal ring. This second testicle had no vas deferens of its own.

It was not found possible to bring down the lower testis into the scrotum, and both were removed, the spermatic cord being divided at the internal ring (*Fig.* 371). The hernial sac was dealt with and a repair was fashioned, using continuous rows of nylon obliterating the inguinal canal. The patient made an uninterrupted recovery.

THE SPECIMEN.—On macroscopical examination there was an area of recent hæmorrhage in the lower testis, well shown in the photograph (*Fig.* 371). The upper organ was normally formed. The absence of a vas deferens was confirmed. Both organs were submitted for section.

Microscopical Appearance (*Fig.* 372).—The upper (abdominal) organ is testicle. The seminiferous tubules are atrophic and scanty. The lower organ is also testis. The section shows that seminiferous tubules are present, but there is no evidence of spermatogenesis.

DISCUSSION

The testicular anomaly has not been constant in the cases described in the literature. Three different types have apparently been found :—

A. A normal testis with a second testis which has no vas deferens or epididymis.

B. A normal testis and a second testis which has an epididymis but no vas deferens.

C. Both testes with epididymes, but the vas deferens of the supernumerary organ continuous with that of the normal one.

Both our cases apparently fall into Group B. *Case* 2 certainly does, but *Case* 1 may possibly be an example of Group C, for a vas deferens from the upper testis may have been present in the thickened fold of tissue connecting the two organs and joining the epididymis of the lower one, as in King's case. Unfortunately this fold, which on palpation contained only vessels, was not sectioned.

In order to explain how polyorchidism with the variations in the vas deferens and epididymis might be produced, it is necessary to dwell briefly on the development of the testis.

The primitive sex gland appears as a thickening of the cœlomic epithelium on the ventromedial side of the mesonephros at about 5–6 weeks (6 mm. embryo). The thickening continues till a definite ridge (the genital ridge) is formed. As this ridge increases in size, grooves appear on its lateral and medial sides so that the tissue between them forms a mesentery—the mesorchium. At about the 13-mm. stage the covering epithelium of the middle two quarters of the ridge sends a number of cords into the underlying mesenchyme. The cephalic and caudal quarters then degenerate, the cephalic one forming the appendix testis, and the caudal one assisting in the formation of the future gubernaculum. At about the 25-mm. stage the epithelial cords are cut off from their parent epithelium by the development of a dense fibrous layer, the future tunica albuginea. The cords, which canalize, become the

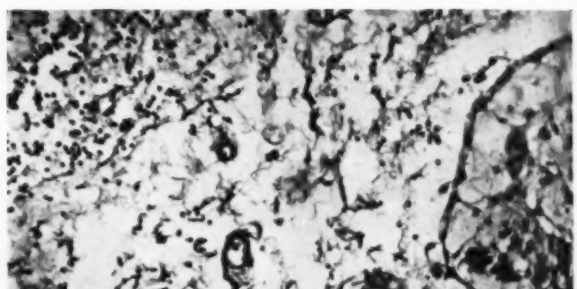

FIG. 372.—*Case* 2. Microscopical appearance. (× 186.)

seminiferous tubules, and grow into the mesorchium, where they form the rete testis and join with the undegenerated tubules (5–12 in number) of the mesonephros. These tubules become convoluted and form the head of the epididymis. They are already, at an earlier stage of development, joined to the mesonephric (Woolfian) duct which forms the rest of the epididymis and the vas deferens.

The generally accepted theories regarding the formation of two testes are that it is due to : (1) Reduplication of the genital ridge. (2) Degeneration of parts of the mesonephric components. (3) Division of the genital ridge by peritoneal bands.

It is easily seen that reduplication of the genital ridge would produce two testes, but if this were the true explanation then both testes should be of the same size, and both should be of normal size. In fact, in the recorded cases where mention is made of size, the normal organ has usually been bigger than the supernumerary one, and the combined bulk has been approximately that of the

unaffected side. In our *Case* 2, however, it was the supernumerary one which was the larger.

We reject the theory that polyorchidism is caused by degeneration of parts of the mesonephric components because we fail to see how this can, in any way, influence the genital ridge itself.

Transverse division of the genital ridge could take place at any part of the ridge, and would, therefore, produce two testes of unequal size. The combined bulk of the testes, however, would only be equal to that of the other side. If the division cuts off a part of the genital ridge which does not come into direct contact with the rete testis, then two masses will be produced, one, probably the larger, connected to the mesonephric components, and having, therefore, in the adult state, testis, epididymis, and vas, while the other will have testicular tissue only. If the division severs the ridge at the point where the testicular components meet the mesonephric components, then two organs will be produced, each of which will have an epididymis. If the division extends to the mesonephros then the upper testis and epididymis will have no vas deferens, while the lower one will. If, on the other hand, the division does not include the mesonephros, then both testes and epididymes will be joined to the one vas (*Fig.* 373).

We feel that this is the only theory that will explain the production of two testes from one sex gland together with the recorded variations of the epididymis and vas deferens. Cases of polyorchidism must be proved by operation and by histological examination of the abnormally placed organ. This is important because, not only are there the obvious diagnostic pitfalls such as cyst of the epididymis, hydrocele of the cord, tuberculosis of the globus major, and the like, but the condition also occurs in which two testes, each with normal testicular sensation, lie in one side of the scrotum. This latter condition has been variously termed ' pseudo-duplication ' and ' transverse ectopia '. One testis crosses the midline during its descent, and joins its fellow in the opposite side of the scrotum. Their vasa may stem from a single seminal vesicle, the common trunk dividing to supply a duct to each testicle, but the testicular arteries always arise normally from the aorta, one crossing the midline to reach the opposite side of the scrotum. Marsh (1911) and Banks (1926) report 2 such cases, and there seems to be strong presumptive evidence that the case reported by Lowe (1911) was one of this variety.

When strict criteria are rigidly applied it becomes evident that the number of reported cases of true polyorchidism is small. It is interesting, therefore, that two cases should present in one hospital within a short period of time. Other considerations, however, cause one to wonder whether the condition is really as rare as has been supposed. In 3 cases (Jordan and Dodson, 1934; Cranz, 1942; and our *Case* 2) the supernumerary organ was intra-abdominal and its discovery at operation has been entirely fortuitous. King (1931) points out that, whereas the remains of a gubernaculum could be clearly distinguished fixing the lower pole of the lower testis to the scrotum in his case, the upper testis had no gubernacular attachment, a state of affairs which one would expect when one examines the embryology of the abnormality. The role of the gubernaculum in the descent of the testis has been the subject of much discussion, but most authorities agree that it acts as a guide, conducting the descending organ to its final destination. One may assume, therefore, that the supernumerary organ, having no such guide, will only pass through the inguinal canal in a small proportion of cases.

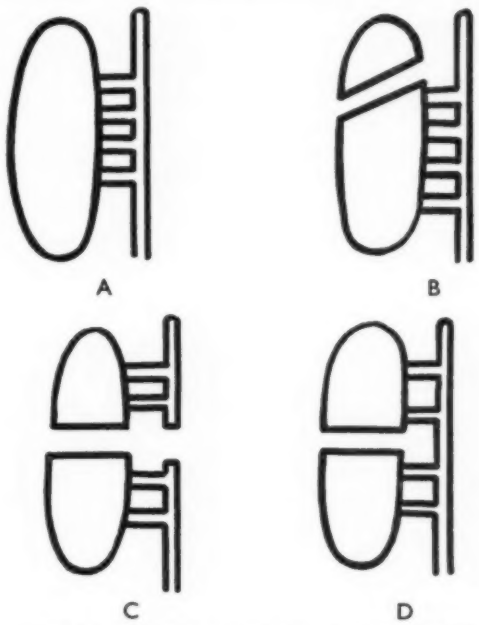

FIG. 373.—Diagram to show the normal relationship of the testis and epididymis and the three possible anomalies. A, To show the middle two quarters of the genital ridge, the mesonephric tubules, and the mesonephric duct. B, Division of the genital ridge with only one part of it connected to the mesonephric components (Group A). C, Division of the genital ridge and the mesonephros (Group B). D, Division of the genital ridge without division of the mesonephros (Group C).

We feel that these facts support the contention than an unknown, but possibly appreciable proportion of the male sex may be the posessors of a third and intra-abdominal testicle, and live their allotted span without the abnormality being discovered. In approximately 75 per cent of reported cases of polyorchidism the abnormality has been left-sided.

Failure of descent alone is more commonly right-sided. A study of the previously reported cases shows that supernumerary testes are commonly undescended. The factors at work in the production of the two conditions—very early developmental abnormalities in the first, and a mixture of mechanical and hormonal aberrations at a later stage in the second—are manifestly different. Again, we may surmise that the fact that the supernumerary testis lacks a gubernaculum may be the reason for the association of the two conditions.

Both our cases were found to have indirect inguinal herniæ, as had other reported cases. However, since 80 per cent of undescended testicles are so complicated, the association in these cases is more likely to be with the imperfect descent than with the polyorchidism.

Boggan points out that spermatogenesis was only recorded in 6 cases before 1933, and comments that

it is only likely to occur in supernumerary organs which have reached the scrotum. There was no evidence of spermatogenesis in the organs submitted to histological examination in the 2 cases reviewed here. It is accepted that spermatogenesis never occurs in an abdominal testis, and that in all cases of maldescent, spermatogenesis is imperfect.

We believe that, in both our cases, the pain complained of was produced by minor episodes of torsion of the testicle. This is borne out by the discovery of a large recent hæmorrhage in the lower testis of *Case* 2, and the histological report on *Case* 1, which also shows areas of recent hæmorrhage. In each case the symptoms were referable to an organ lying in the inguinal canal.

No case of malignant disease of a supernumerary testis has yet been recorded, though Goldstein and Cassilli (1939) reported a case of rhabdomyosarcoma of the cremaster muscle in a case of polyorchidism.

It seems to us that the apparent complications of polyorchidism are, in reality, merely the well-known complications of imperfect descent of the testis, and it is unlikely that the possession of three testicles need cause their owner any disquiet, providing all three are safely housed in the scrotum, even if the claims of an earlier generation regarding the prolificacy of one so endowed have not been borne out in the light of more recent knowledge. If, however, the supernumerary organ is not in the scrotum, operation should be advised because, by virtue of its position, it is probably associated with a hernial sac, and it is liable to be complicated by trauma, torsion, and possibly malignant disease also.

Only one previous case of torsion associated with polyorchidism has been reported (Boggan, 1933). Although our cases were associated with imperfect descent, in his case all three testes were in the scrotum. We have, therefore, examined the known factors which predispose to torsion, in an endeavour to determine whether any of them may operate in cases of duplication of the testicle. Muschat (1932) states that torsion may be intravaginal or extravaginal. The first type can only occur when there is complete investment of the testis, epididymis, and a section of the spermatic cord by the tunica vaginalis. Both cases reported here were associated with a hernial sac, but in neither case was the torted testicle invested by the peritoneum of the sac. When no hernial sac is present one would expect to find that the lower testis has a tunica vaginalis and that the supernumerary organ has none. This was the finding in the case reported by Handley and Crawford (1944).

Extravaginal torsion is only accepted with reservations by most authors, and Young and Davis (1926) state that it occurs only when a severe external force tears the undescended testis from its scrotal attachments. Meltzer, quoted by Muschat, lists abnormalities which have been described in cases of torsion of the testis. He includes absence of the gubernaculum, absence of the normal attachment between the posteromedial surface of the epididymis and the inner wall of the scrotum, and the presence of loose and mobile connexions between testis and epididymis. We have pointed out that, on developmental grounds, one would not expect a supernumerary testis to possess a gubernaculum, and obviously a testis which is supernumerary and undescended will have no other connexion with the scrotum. Abnormalities such as a well-developed mesorchium are surely to be found more commonly in ill-formed supernumerary testicles than in the normal organ.

From a study of the mechanism of production of intravaginal torsion one realizes that the twist is complete and irreversible, whereas extravaginal torsion is likely to be much less complete because an organ not entirely pedicled cannot rotate to any great extent. This hypothesis is supported by the findings in the two cases presented here as, in both of them, infarction had been on a small scale, and gross interference with the blood-supply had obviously not occurred.

Extravaginal torsion is, therefore, the type which one may well expect to find complicating a case of supernumerary testis associated with undescent, and it is rather surprising that torsion in a case of polyorchidism has appeared in the literature only once previously.

SUMMARY

1. The previous literature on the subject is briefly reviewed.

2. Two cases of polyorchidism associated with torsion of the testis are reported.

3. Theories of production of the abnormality are discussed.

4. It is suggested the polyorchidism may not be as rare as has been supposed hitherto.

5. The relationship between polyorchidism and imperfect descent is discussed.

6. It is pointed out that operative treatment is only necessary *if* there is imperfect descent.

7. Some factors which may produce torsion are discussed, and it is shown that torsion in this type of case will never be complete and overwhelming.

For permission to record these cases we are indebted to Mr. J. B. Oldham and Mr. Maurice Silverstone, under whose care the patients were admitted to hospital.

We should also like to thank Mr. Oldham for his encouragement and help in the preparation of this paper.

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THE AETIOLOGY OF PILONIDAL SINUSES

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THE present conception of the nature of pilonidal sinuses held by most surgeons and teachers of surgery is that a canal present in the embryo is only partly obliterated in the post-anal region, leaving a pit opening on to the skin. This attitude has resulted in the grouping together of many different conditions seen in this region under the term of 'pilonidal sinus' in the belief that they all have this common origin. The confusion of thought which can follow the grouping of many different conditions under this term is seen in a recent review by Turell and Gladstone (1951). These conditions include the post-anal dimple, congenital dermal sinus, post-anal dermoid, and even the sacrococcygeal dermoid occurring on the anterior aspect of the coccyx. Because it is possible that these lesions do in fact arise from a defect such as that postulated by Walker and Bucy (1934), such a grouping is worth retaining provided the pilonidal sinus is not included.

It is unusual to find hair present in these conditions, whereas in the pilonidal sinus it is the rule. This observation, with the probability that no developmental basis exists for the formation of the true pilonidal sinus, makes it important to classify the pilonidal sinus as separate from other conditions in this region. It seems that because the pilonidal sinus is the commonest lesion seen in the post-anal region, its name has often been used carelessly as a general term to embrace that group including the post-anal dimple, post-anal dermoid, and congenital dermal sinus. The object of this paper is to establish the fundamental distinction which exists between those rare developmental lesions in the coccygeal region and the common lesion known as the pilonidal sinus.

Definition.—A pilonidal sinus is a cavity in the subcutaneous tissues which is lined by granulation tissue, contains hair, and communicates with the surface by a track line usually by squamous epithelium continuous with the epidermis.

The usual site for a pilonidal sinus is in the midline between the buttocks. However, identical lesions occurring in other sites must be considered along with those in the post-anal region.

Past Theories of Origin.—Until recently most theories of origin were based on the embryology of the caudal end of the neural groove.

1. Persistence of the neurenteric canal. How such a preposterous suggestion could have arisen is incredible, but nevertheless it has at times been thoughtlessly claimed as the origin of the post-anal sinus. Needless to say, any persistence of this canal would result in a fistula from the bowel to the lumbar region of the spinal cord (Keith, 1948).

2. Failure of the medullary lips of neural canal to close completely at the posterior end of the neural groove (Mallory, 1892). Such an abnormality does occur, but is recognized in varying degrees from spina bifida occulta to myelocele.

Another rare form of this embryological defect has been shown by Shenkin, Hunt, and Horn (1944) to occur where a sinus from the skin over the sacrococcygeal region communicates by a cystic filum terminale with the central canal of the spinal cord. It is clear that these defects have little in common with the pilonidal sinus.

3. After fusion of the neural crest, this fails to separate from the cutaneous epithelial layer, which is converted into a deep pit at this site by growth of the adjacent surrounding soft tissues. The lesion is an epithelial sinus from the skin in the midline of the back to the meninges of the central nervous system. This important connexion clearly distinguishes this lesion from the pilonidal sinus. The recent review by Cardell and Laurence (1951) of this condition known as congenital dermal sinus, suggests the relation of the deep dermoid cysts in this region to the dermal sinuses.

4. The novel theory that the pilonidal sinus is an acquired lesion was introduced by Patey and Scarff (1946), and independently by King (1947). These writers discarded the developmental basis of these lesions and demonstrated features in support of the view that they originated by penetration of loose hairs from the exterior into the subcutaneous tissues.

Some aspects of the pathology will be described in this paper to support the acquired nature of the pilonidal sinus.

OBSERVATIONS

Incidence.—The type of person in whom the pilonidal sinus occurs is of importance in determining its aetiology.

1. *Age.*—The youngest of a series of 40 patients treated recently was 17 years and the eldest 49 years. Most patients are about 20 years old.

2. *Sex.*—It was found that males predominated in the proportion of 3 to 1 in a series of 40 patients. This, however, shows that females are rather more often affected than had previously been thought.

3. *Skin Type.*—This will be seen to be of great importance in determining the occurrence of a pilonidal sinus. The type of skin in which pilonidal sinuses are commonest is that which produces dense stiff dark hairs, and usually is greasy through seborrhœic gland activity. It is very uncommon to find a sinus in a fair-haired person unless the hair is

of the stiff, often wavy, type. The freedom from sinuses of the soft-skinned and silky, fair-haired persons is most likely attributable to two factors the significance of which will be discussed later: First, the texture of the hair is soft and pliable, and it will be seen that stiff bristly hair is that usually

FIG. 374.—Operation specimen from a young male patient of Italian parentage. The midline crypts have been laid open to show their communication with the deep cavity containing the tightly packed tuft of crisp black hairs. These hairs were lying quite loosely in the cavity. Note the prominence of the hair follicles, and the secondary lateral opening to the left of the midline.

found in the sinuses; secondly, the openings of the hair follicles are smaller and this must be related to less active sebaceous gland activity in this type of person.

4. *Occupation.*—This often has no obvious relation to the occurrence of a sinus, but those subjecting the sacrococcygeal region to repeated minor trauma as in sedentary occupations appear to be more often afflicted. This factor was more strikingly demonstrated during the war, when the riding in Army vehicles was blamed for the condition known as 'jeep seat'.

Sites.—Lesions similar in all respects to the common post-anal pilonidal sinus may occur elsewhere in the body. Smith (1948) reports some pilonidal sinuses in the perineum, King (1947) in the horizontal gluteal fold on the buttock, and the writer has recently observed a sinus one inch off the midline, high in the lumbar region, of a barber.

The most significant site for true pilonidal sinuses to be found is in the interdigital cleft of the hand. It will be seen later that we have in this lesion evidence provided by a 'natural experiment' which proves the acquired nature of pilonidal sinuses.

The pathology and aetiology of the interdigital pilonidal sinus have been discussed by King (1949) and Hueston (1952). Usually this condition is seen only in barbers and is due to the intrusion of hair fragments, cut from the customer's head, migrating up the fingers and penetrating the soft skin of the web space. A similar condition has been reported in a wool-shearer by Matheson (1951) and in the hands of milkers by Becker (1930). Here there was penetration of the skin of the web space by fragments of wool in Matheson's case, and by the small stiff hairs from the cow's udder in Becker's cases.

Operative Findings.—It is difficult to believe that some of the observations possible at operation have not raised more doubts in the simple embryological explanations of this condition.

1. The sinus openings: There are usually two or more small pits in the midline over the sinus cavity. It is uncommon to see a hair protruding from one of these pits. In 19 of a series of 40 cases there was also a secondary lateral opening (*Fig.* 374) from which pus was discharged during acute inflammatory episodes. There is a belief, apparently due to Oldham (1946), that these secondary lateral openings are commoner on the left side, but it seems that this view requires revision, for in the 19 cases mentioned earlier there were 9 openings on the left side, 9 on the right side and 1 case had an opening on each side of the midline.

2. Injection of methylene blue or indigo carmine into an opening is used frequently to allow accurate and complete excision of the deep sinus. This technique usually shows that the pits are in communication with the sinus because injection through a lateral opening causes a flow of dye from the midline. However, a deep sinus loculus is sometimes found not to have been stained by the dye and it seems that its use may be misleading. Complete excision of all ramifications can be achieved by relying on the typical appearance of the granulation tissue, the surrounding fibrosis and the visible hair fragments.

3. The state of the hairs: The colour of hairs found is that of the external hairs. Short hairs are the rule within these sinuses, usually less than one inch in length, and usually coarse and stiff. The hairs are often present in great numbers, when they assume the appearance of a thick tuft as in *Fig.* 374. In other cases the hairs are found in smaller numbers and scattered in the soft tissues as well as in the sinus cavity.

The most significant single observation to be made on the hairs is that they are not growing in the sinus. The hairs are either lying loose within the cavity of the sinus or embedded in the granulation tissue of the wall. Loose hairs are seen in *Fig.* 375, and the mass of hairs seen in *Fig.* 374 was quite unattached within the sinus cavity. Close examination of the hairs present reveals that a hair follicle is not often attached, and when a follicle is present it is found on the outer or free end of the hair. When the ends of hair fragments present in the sections are examined one may find the abrupt appearance shown in *Fig.* 376, where clearly the hair has been fractured and is not growing. In the extreme depth of a sinus the ends of the hairs closest to the sinus wall are often squared (*Fig.* 377). No chance tangential section can produce this appearance

at the end of a hair, and the only conclusion is that these hairs are lying with their clipped free end innermost.

Such observations should convince the most sceptical that these hairs present in a pilonidal sinus are not growing from the wall of the sinus.

4. The relation to previous scars: some cases of pilonidal sinus appear to have originated at the

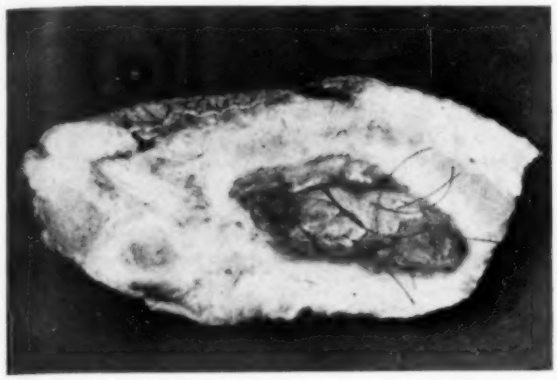

FIG. 375.—Another operation specimen divided in the midline and showing short hairs lying loosely in the deep abscess cavity.

site of a previous scar. The long subcutaneous sinus seen in *Fig.* 378 developed in the five months following excision of a true fistula-in-ano. Its external opening is at the upper end of the operative scar. The probability here is that hairs began to enter the subcutaneous tissues through the wound during the post-operative phase of granulation, or through the soft newly formed scar. Foreign bodies of this kind in the subcutaneous tissue would migrate owing to the frequent and constant movement of the skin

FIG. 377.—Section of the deepest part of an interdigital pilonidal sinus on a barber's hand, showing the abrupt square end on several hair fragments. This is the clipped free end of a hair and indicates that the hair has entered the sinus with its free end leading.

of this region over the deeper tissues and indeed the length of the sinus itself indicates such a mechanical factor.

The recurrence of a pilonidal sinus after apparently complete excision and healing need not be regarded by the surgeon as evidence of failure to excise the previous lesion completely. Persistence of the same local conditions, particularly with the presence of a depressed scar where hairs may penetrate readily accounts for most recurrences. The external opening in such recurrent sinuses is usually found near the end of the scar.

FIG. 376.—Section from the wall of the deep cavity in the specimen shown in *Fig.* 374. The free surface is to the left and consists of granulation tissue. Note a fragment of hair lying embedded in the wall, and showing an abrupt end.

Histology.—The structure of a pilonidal sinus is a track lined in its outer part by stratified squamous epithelium and usually in the depths by granulation tissue surrounding a collection of hairs. The stratified squamous epithelium lining the main sinus

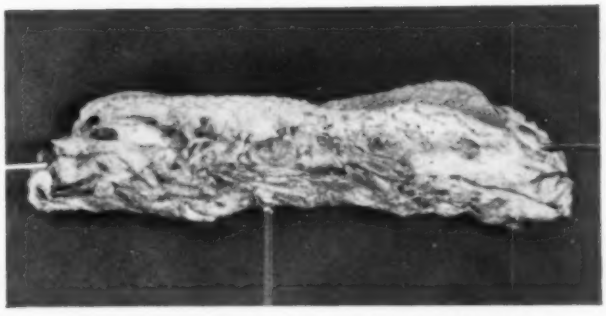

FIG. 378.—Operation specimen with metal probe in place to show the subcutaneous course of a pilonidal sinus which followed excision of a fistula-in-ano. The external opening is to the right and was 3 cm. from the anal margin, at the site of the scar from the excision of the fistula. Several loose hairs were present in the deep part of the sinus to the left of this specimen. Total length of sinus was 8·5 cm.

track is continuous with the adjacent epidermis. In the depths of the sinus this epithelial layer thins out and finally disappears. The cavity containing the hairs is lined by granulation tissue of moderate vascularity. A significant observation is the presence of hair fragments within this granulation tissue. These hair fragments are surrounded by the low-grade inflammatory response seen throughout the wall, with some orientation of the connective-tissue

elements around them (*Fig.* 376) and in places giant cells of the foreign-body type can be seen adjacent to the hair (*Fig.* 379). No hair follicle can be seen on histological section. The orientation of giant cells around a hair fragment may possibly lead to confusion with a hair follicle, as clearly occurred in Fig. 5 of the paper by Davies and Starr (1945).

FIG. 379.—Section showing hair fragment embedded in the granulation tissue forming the wall of the sinus. Note the orientation of giant cells about the fragment, giving an appearance which might lead to confusion with a true hair follicle.

DISCUSSION

The successful treatment of any condition usually only follows an understanding of its aetiology and pathology. A failure to recognize the nature and origin of the pilonidal sinus seems to be a reason for the great variety of surgical treatments recommended. It is, therefore, of importance to establish that the true nature of the pilonidal sinus is an acquired lesion of the foreign-body granuloma type resulting in a chronic sinus, and not related to those developmental errors occurring in the sacrococcygeal region.

Considering first the age incidence of the lesion, we find that it shows a distribution typical of an acquired lesion (*Fig.* 380 B). The condition is very rare below 15 years and above 50 years, and has its highest incidence at 20 to 25 years. This age distribution differs from that of a true embryological defect, for example congenital heart disease, where the highest incidence clinically is at birth and steadily falls during infancy and childhood, to be much less common in adults (*Fig.* 380 A). If the pilonidal sinus were a developmental defect it would be present at birth. The reason usually given for its late recognition is its failure to cause symptoms until friction or repeated minor trauma leads to obstruction to the orifice and infection occurs. Even if this were so, the sinus should be visible in children and infants; but this is not possible. Those pits and sinuses occasionally seen in infants rarely contain hair, and often have some connexion to the dura or filum terminale. These lesions are clearly embryological defects and not true pilonidal sinuses.

If the pilonidal sinus is not a developmental defect then how is it produced?

It is now believed that the fully developed pilonidal sinus is due to the repeated intrusion of hairs from the exterior. Secondary infection and deep abscess formation accompany the presence of these foreign bodies. The main factors responsible for the initiation of this process are clearly going to be the physical properties of the hairs and the state of the skin allowing penetration by the hairs.

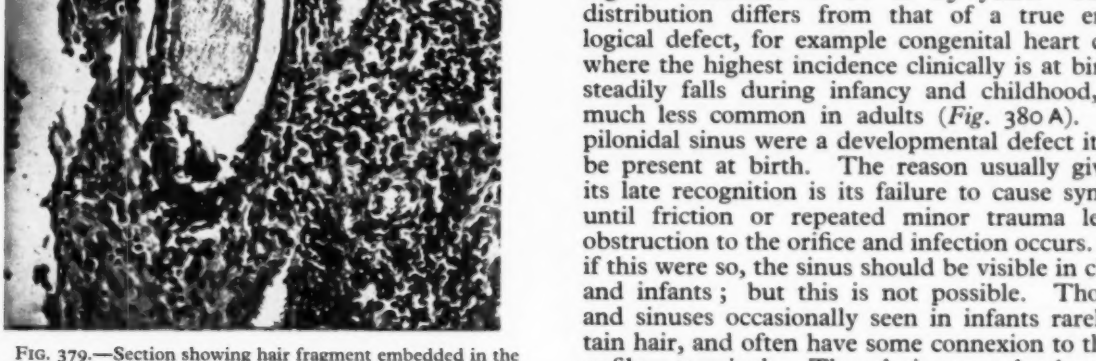

FIG. 380.—A, Age incidence of a congenital lesion. This is the general form of curve given by conditions such as congenital heart disease or Meckel's diverticulum. There is a decrease in the number of cases with each decade. B, Age incidence of an acquired condition. This is the kind of curve found in a condition such as hydatid disease, where the acquired nature of the lesion is undoubted. The age distribution of the true pilonidal sinus gives this type of curve and not that of congenital lesions.

The histological structure of the post-anal pilonidal sinus is in every respect similar to that of the interdigital pilonidal sinus of barbers (Hueston, 1952) and wool-shearers (Matheson, 1951). The conclusion from a consideration of the histological appearances alone is that this condition is an example of a foreign-body sinus due to the presence of hair fragments.

The reason why pilonidal sinuses are seen in dark wiry-haired individuals is almost certainly that the hairs possess greater rigidity so as to be less easily bent and folded than the soft hair of fair people. This facilitates penetration of skin and sinuses.

Since the rigidity of a hair is a function of its cross-sectional area and its length, then the shorter the hair the greater the rigidity, and the readier the penetration. This would explain the finding of short stout hairs within sinuses and not the long soft coiled hairs often seen in true dermoid cysts where the hair has quite clearly been locally formed.

The skin of dark, hairy people possesses larger hair follicles with greater seborrhœic activity than that of fair persons. These follicles are largest in the region of the natal cleft, and are very obvious in the specimen shown in *Fig.* 374. It seems that the accumulation of sweat and epithelial debris in the natal cleft may gain entry to some of these large hair follicles, particularly when repeated friction or minor trauma is applied to the part and in the hot climates where skin gland activity is greatest. Enlargement of those follicles is the result, and usually occurs in the midline, but, as was mentioned earlier, may occur in the perineum or the horizontal gluteal fold.

The short hairs growing on the buttocks and in the natal cleft of hairy persons are shed at intervals and tend to collect in the natal cleft and perineal region unless meticulous care is taken with toilet. Hairs from the neck and skin over the back when shed will lie deep to a shirt or underclothes and find their way to the same region, along with the smaller epithelial fragments of these parts. By the movements of the opposing surfaces of the natal cleft some of these short hairs will be forced into an enlarged hair follicle. If a hair follicle is still retained on a loose hair, the sharper end will naturally be the one to enter the tiny crypt first, and this corresponds with the operative finding that hair follicles when present are directed out toward the surface opening of the sinus. When a hair is seen projecting from the orifice of a pilonidal sinus it is easily extracted, because it is in the process of entering the sinus and not growing out from it.

It is now recognized that the growth of skin elements, including the hair follicles and sebaceous glands, is under endocrine influence, particularly that of the androgens, and the advent of puberty with the subesquent increase in the activity of these elements is probably of importance in determining the age incidence of this condition.

Although in the post-anal region it is most likely that the enlarged follicles are of importance in allowing the entry of foreign hairs, the presence of such a portal of entry is not essential for the formation of a pilonidal sinus. This is shown in the interdigital pilonidal sinus of barbers, where the short stiff hair fragments from the customer's head are able to penetrate the intact skin of the interdigital web space where there are no hair follicles. Barbers recognize the readiness with which hair fragments can penetrate the conjunctiva, and the penetration of the intact skin on the hands is aided by the softening due to frequent applications of oily hair lotions. The same factor of skin softening, from the collection of skin secretions deep in the natal cleft, may be a factor in determining that the midline hair follicles are those most commonly affected.

Once the skin opening has been formed there is ready access to any hairs and epithelial debris which may collect in the natal cleft. Secondary infection probably occurs early in this process, and leads to a slow progressive extension of the deeper part of the crypt. However, when the infection extends beyond the lining wall of the cavity, which is most likely when the sinus track is obstructed, the surrounding cellulitis attracts the attention of the patient. Suppuration is then most likely with deep abscess formation. Lateral secondary openings from these deep abscesses are due to the less dense structure of the subcutaneous fat on either side of the midline.

The persistence of a pilonidal sinus is due to the retained hairs and the epithelization of the main track which is a general feature of chronic sinus formation. On the other hand, an appreciation of the mode of formation of this troublesome condition should lead to a lessened incidence by stressing the importance of personal toilet in hot climates and in people of the type likely to be afflicted. There are distressing cases in which a recurrence of a pilonidal sinus occurs after a complete excision of all sinus ramifications. These are due to the persistence of the determining factors mentioned. Thus prevention of recurrence includes careful toilet of the region.

SUMMARY

1. The pilonidal sinus is an acquired condition and is to be segregated from the rarer developmental abnormalities seen at the lower end of the spinal column.

2. The pilonidal sinus is a chronic sinus of the foreign-body type due to the penetration by short stiff hairs into the subcutaneous tissues. Extension occurs by persistent low-grade infection.

3. Enlarged hair follicles in the midline are the original portals of entry for the hairs, and the age incidence of the lesions is possibly related to the increased activity of these skin elements after puberty.

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CONCERNING THE STUDY OF THE PHYSIOLOGY OF PATIENTS SUFFERING TRAUMA: A METHOD OF WEIGHING THE PATIENT

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HAVING found an accurate and easily obtainable system of weighing patients undergoing operations, I feel that it may be of assistance to others who are interested in the same problem and its applications.

Since the latter part of the war, increasing interest has stimulated attempts to devise accurate ways and means of measuring any change of the water content of the body during an observed period of disease or treatment. If it is decided that measurement of change in weight is to be used to give evidence of variations in water content, obstacles immediately present themselves.

For example: In order to measure the loss of water by a patient under treatment with digitalis for cardiac failure, it may be considered necessary, besides measuring the urine output, to weigh the patient. By virtue of the condition it is imperative that the patient be weighed in bed with all the bedclothes. To do this day after day in the same conditions of humidity, besides knowing the exact weight of the sheets and blankets that are often being changed, demands the facilities of a special small ward set aside for metabolic study. This luxury, which is perhaps not yet attained in this country, enables the observer to make observations which are as accurate as they can be. Changes in temperature and humidity cause appreciable day-to-day variation in the weight of bedclothes, pillows, and mattress, added to which is the difference in weight of clean sheets, pillow cases, etc. One has only to hold one's clothes to the fire a few days after they have been washed, dried, ironed, and 'aired' to appreciate the vast amount of water that has re-entered the material and therefore the weight gained!

From a surgeon's point of view, whether traumatic, burns, chest, plastic, or 'general', studies in the change of weight associated with measurements of blood-volume, electrolytes, and so on may have great practical importance. The surgeon is in a more fortunate position than the physician in that he always has the opportunity of studying the patient in a special room where humidity and temperature may be kept steady. This room is the operating theatre and the period that the patient may be in it, together with the occurrence of an operative procedure, furnishes the surgeon with the opportunity of studying his patient and measuring body changes that occur. By knowing how to correct these changes it may be that he will advance a little further on the road of 'making surgery safe for the patient'.

Changes in humidity and temperature are small in theatres with air conditioning and when clothing is at a minimum, and if conditions are as suitable as this, all that remains before study can commence is the discovery of a means of weighing the patient before and after the operation. A few years ago in the Queen Elizabeth Hospital, Birmingham, efforts were made to measure blood-loss by weighing swabs before and after use, and such a method of measurement is still being used in this locality. The method entails the use of dry swabs and the immediate weighing of these after being suitably blood-stained; but not all the blood lost or destroyed can be calculated in this way and, in addition, evaporation occurs and errors are multiplied, so that at best it is only one facet of the whole rather gigantic and difficult problem.

On contact with a well-known company who manufacture weighing machines (Messrs. W. & T. Avery Ltd.) it was found that they are able to construct a trolley with a weighing machine incorporated, and at first sight this might be thought suitable. However, it is the practice in the Queen Elizabeth Hospital to transfer patients from the operating table directly to their beds, thus removing the necessity of an additional move in the ward, away from the watchful eye of the anæsthetist, with the risk of lowering blood-pressure and impairment of condition. The use of any trolley method tends to be against all interest in the patient's welfare.

The best method is to weigh the patient in the theatre on the operating table, and it is with the technical advice of Messrs. W. & T. Avery Ltd., and at a cost of several shillings, that I have constructed a weighing system which I find is accurate enough to measure variations of one ounce. The following is an account of the apparatus and the method.

Apparatus.—

Two "Avery" weighing machines (chair type).
Two wooden yokes to rest on chairs and to support stretcher poles.
Two ash stretcher poles.
Two cross-connecting rods for the poles.
Four sections of tubular steel (gas main 2 in.) each 2 ft. long, to provide extensions for ash poles.
One canvas stretcher capable of taking the ash poles.

Assembly and Method.—With the stretcher beneath the patient the poles are threaded into position. The patient is lifted on to the operating table and the poles are left in the stretcher, length being given to them by sliding on the metal extension pieces by a constant amount (this being marked by a screw or line drawn on the wood). The cross-connecting rods are passed along the pole handles to stabilize the stretcher. The operating table is 'pumped up' as high as it will go and the weighing chairs with the wooden yokes on their arms wheeled into position directly beneath the metal extensions of the poles. The height of each yoke should be sufficient to take the extended stretcher poles when the operating table is pumped as high as possible, so that there is enough clearance between the stretcher

and the table when lowered. The parts of the yoke which take the weight are padded with felt.

With the chairs and yokes in the position described it is necessary to make sure that the chair arms are free from the vertical path taken by each end of the table. If this is satisfactory, the release pedal on the operating table is depressed and the table allowed to subside slowly as far as it will go. The patient

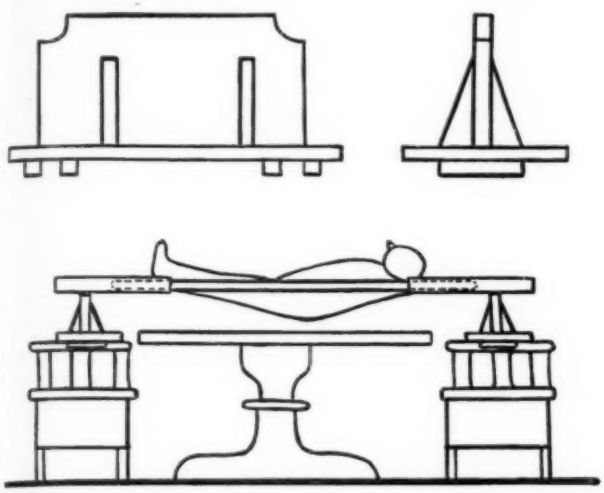

FIG. 381.—Above are shown two perspectives of the wooden yoke. Below is a drawing showing the patient in the weighing position between the two chair weighing machines after the operating table has descended.

by this time is suspended on the stretcher in mid air between the two weighing machines. The corrugated tubing connecting the patient's airway to the anæsthetic machine is disconnected for sufficient time (about 60 sec.) to enable the weight to be measured. This is done by adjusting each machine until the weighing beams are stationary at the neutral point and then the amount on each is recorded. There is no undue vacillation of the beams as the weight is virtually 'dead weight', a great advantage of this method. Three readings are taken, in between which the weight is taken off the machines by pumping up the table.

After the pre-operative weighing, the table is 'pumped up' to allow the removal of the yokes and machines, and then the table is allowed to subside to a level convenient for the dictates of the operation. The whole procedure is repeated at the end of the operation, each weighing machine being placed as before. (*Fig.* 381.)

Example.—

Name: Mr. D——.
Date: Feb. 18, 1952
Temp. of theatre: 75–76° F.
Humidity: 40–42 per cent.
Operation: Partial gastrectomy.
Diagnosis: Duodenal ulcer.
Time: 1250–1410 hr.

Total Weight:

	Feet st.	lb.	oz.	*Head* st.	lb.	oz.	
Before:	6	11	1	7	2	3	88565·5 g.
After:	6	9	2	7	2	9	87846·5 g.
Plus or Minus	− 0	1	15	+ 0	0	6	− 719·0 g.

Total Weight Change: Minus 719·0 g.

Adjustments for measuring True Change in Body-weight:

Weight of materials added to body:

Intravenous drugs	22 g.	
Intravenous drip	Nil	
Ligatures and dressings	56 g.	
	—	78 g.
Change in weight of operating gown, socks, and pillows		Nil

True Change = Minus 797 g. (719 + 78).

Adjustment for measuring True Change in Body-weight excluding Tissues excised and Fluids aspirated, etc.:

Weight of tissues, etc., removed from body:

Tissue—stomach	113 g.	
Aspiration	28 g.	
Water absorber	14 g.	
Blood from arm	10 g.	
	—	165 g.

= Minus 632 g. (797–165.)

This weight (632 g.) is the total weight of blood, plasma, and water lost by the patient. This figure will tend to be on the low side unless the abdominal cavity is rendered completely free of any liberated blood. The ways and means whereby the actual amounts of these fluids are measured are not included in the scope of this paper on the method of weighing, but they are sign-posted by blood-volume estimations.

From experience of the method another interesting problem is already presenting itself for study. This is the difference in the change in weight at each end of the stretcher. In the example given, the head end gained 6 oz. and the foot end lost 1 lb. 15 oz. The patient was not moved during the operation and the weighing machines were placed in the same position referable to the end of the poles. This may indicate a shift of the centre of gravity towards the head consequent upon the decrease in weight of the lower limbs due to vasoconstriction. As the patients have been in bed for at least eighteen hours prior to operation it is not due to the change from the upright to supine position.

My thanks are due to Professor F. A. R. Stammers for his encouragement; Messrs. W. & T. Avery Ltd. for their technical advice; and Mr. N. Crawford, Technician to the Department of Surgery, for his assistance in the construction of the apparatus.

A NEURILEMMOMA OF THE PELVIS PRESENTING WITH ACUTE MANIFESTATIONS

By HENRY B. YOUNG

SENIOR SURGICAL REGISTRAR, WOLVERHAMPTON GROUP OF HOSPITALS

THE specific nerve-sheath tumour or neurilemmoma is a growth of very infrequent occurrence. One of the most complete surveys of this tumour was made by Arthur Purdy Stout and his colleague Wayne Carson, and recorded in the *American Journal of Cancer* of August, 1935. It dealt with some 246 cases from a world-wide collection, and states the site clearly in all. The abdomen was one of the rarer sites and the pelvis rarer still. They quote 6 cases by different people: (1) Erb's (1924)—attached to the obturator nerve, projecting into the obturator canal and below Poupart's ligament. (2) Frank's case (1934), which was attached to branches of the sacral plexus, and mistaken clinically for ectopic gestation. (3) Krekeler's case (1924), which was also described by Krumbein (1925), in which the tumour lay behind the rectum, projecting into it. (4) Moreau and van Bogaert's (1923), which was attached to the crural nerve in the pelvis. (5) Pana's (1931), which lay behind the bladder and prostate. (6) Cases which developed in sheaths of the retroperitoneal and pelvic nerves (Eichhoff (Korbsch), 1932; Pescatori, 1929). Pain was not found to be a striking feature in the tumours, being present in only 13 out of 50. Tenderness was elicited in 19 out of 50. Motor disturbances were definitely rare, apart from occasional complaints of weakness with or without paræsthesiæ.

The case outlined below is of interest because of its rarity, its acute symptomatology, and its motor accompaniments.

CASE RECORD

An otherwise fit man of 24 complained of lower abdominal pain for two weeks, vomiting, especially after food, for three days, a greenish diarrhœa for two days, and a marked increase in the severity of the pain for the last day, which was then also continuous. On the morning of admission a swelling had become noticeable in the lower abdomen, especially the left side.

His past history was one slight attack of dysentery on the way out to Egypt in 1940 and one attack of malaria in Palestine, both illnesses of very transient nature.

ON EXAMINATION.—He looked pale, gaunt, and ill. T. 101°, P. 94, R. 22.

The abdomen showed a large, firm, non-resonant, very tender, fixed but very hemispherical mass in the hypogastric region, extending into the left iliac fossa low down (*Fig.* 382).

Rectal examination showed a bulging, tender mass anteriorly in the pelvis. W.B.C. 8000. Catheterization hardly altered the swelling and the urine was clear. It was decided that the pain and tenderness with diarrhœa and fever made an inflammatory mass the likely diagnosis, although the W.B.C. count was low for an illness of such severity due to this cause. Torsion of some other content, normal or abnormal, was considered a possibility.

OPERATION (H. B. Y.).—This was mainly exploratory. A bimanual examination under the anæsthetic was made first, with little help except to reveal that the total mass was about 9 in. in diameter. A small, low, left paramedian incision was then made, disclosing a blue-black, nodular, œdematous, and vaguely fluctuating mass, lying between the cæcum and pelvic colon, both of which were closely applied to it, excluding the appendix from sight, somewhere within it. The bladder was free from it and a hand could be passed behind the upper two-thirds of it. No pus could be found on digital exploration. It seemed

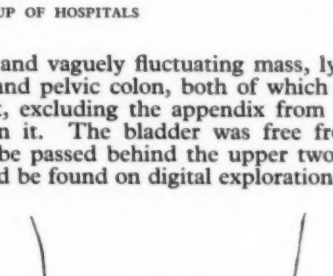

FIG. 382.—Outline of the abdominal swelling. Presented by the patient at the original examination.

to be a closed abscess pointing below. A rectal approach was therefore made and sinus forceps inserted into the bulging mass, producing some sero-sanguineous fluid, but no pus. The impression was that it was either an acute inflammatory mass, possibly appendiceal in origin, with no pus as yet found, or a chronic granuloma, just possibly of amœbic origin, in view of his attack of dysentery, although this seemed likely to have been bacterial. He was given, therefore, sulphonamide, streptomycin, suction, and intravenous therapy. Under this régime his T.P.R. continued to swing markedly, and the leucocyte count varied between 14,000 and 25,000. The swelling waxed and waned, urinary retention occurred intermittently, and pus, mucous, and a little blood were passed per rectum. X-ray, biochemical, and bacteriological examinations were of no diagnostic help. He lost a great amount of flesh, but slowly and steadily the mass subsided—at five weeks to be just palpable above the pubis but solidly present on rectal examination. Sigmoidoscopy showed a small granulomatous area on the anterior rectal wall, probably the site of forceps puncture. However, a grain of emetine was commenced—daily for seven days, and after the fifth day the swelling was found to be considerably smaller but more markedly softer, the role of the emetine in the change being doubtful.

Side by side with this, gradual resolution and general improvement was another surprising inexplicable train of symptoms. His own statement of the sequence of events was that on the third post-operative day he ate some steamed fish which gave him a little abdominal pain, followed by numbness on the outer side of the left leg, after which he found he could not move his foot. Examination showed the presence of 'foot-drop' and diminution of sensation on the middle third of the lateral aspect of the leg and on the dorsum between the first and second toes. Repeated clinical and electrical examinations

established the presence of paresis followed by the reaction of degeneration in tibialis anticus, extensor hallux longus, and tibialis posticus, with transient weakness only in the extensor longus digitorum. The involvement of the posterior muscle pointed to the plexus as the site of the nerve lesion. Was the plexus directly involved in a spreading inflammatory process, was it the object of pressure by the mass, or was the tumour really a sarcoma extending into it? All seemed unsatisfactory explanations.

After seven weeks in hospital he was transferred to a convalescent home, wearing a toe-raising spring.

He was seen as an out-patient from time to time over the next seven months, all the time gaining in health and strength, until he looked and felt 100 per cent fit, only two abnormalities remaining—the firm swelling per rectum and some degree of drop-foot, the tibialis anticus remaining wasted, the other muscles having recovered the power to work against gravity. He could walk without the support for short distances. He was then re-admitted for further exploration.

SECOND OPERATION (H. B. Y.)—The old scar was excised, the wound reopened and enlarged. Many adhesions were found but the cæcum and appendix were normal. The true pelvis was filled with a nodular, solid, yet cystic feeling mass in front of the rectum, but with the sigmoid looped over and firmly adherent to it—all being covered with large vascular adhesions from the omentum. The adhesions were separated, the colon dissected free (one small hole being repaired), the tumour scooped out of the pelvis with the hand, tearing through the fine stretched layer of peritoneum at the base, without encountering any obstructing or anchoring bands of note. The raw area on the floor of the pelvis was then closed and the appendix removed.

There was no appreciable change in the neurological signs on leaving hospital some three weeks later.

PATHOLOGICAL REPORT on the excised tumour (*Fig.* 383) made by Dr. David Hewspear, Senior Assistant Pathologist, was as follows:—

Macroscopical: "An oval encapsulated tumour, fleshy in colour, measuring 8 × 6 × 4 cm. The cut

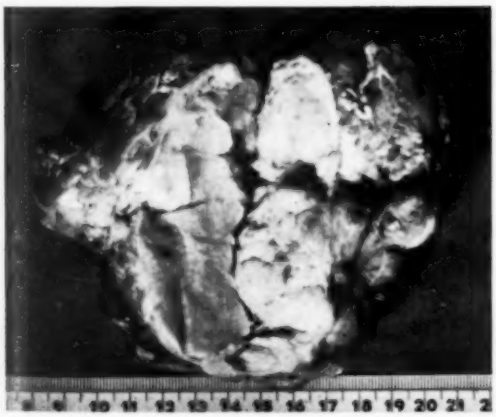

FIG. 383.—Photograph of the tumour removed seven months after the acute phase.

surface showed a whorled appearance and there appeared to be some areas of softening and old hæmorrhage towards the centre of the growth. Sections were taken of the periphery and centre and stained with hæmatoxylin and eosin, van Gieson, phosphotungstic acid, and reticulin.

Microscopical: "The histological picture was that of a typical nerve-sheath tumour or neurilemmoma and consisted mostly of Type A tissue, i.e., cells arranged in an orderly manner giving the well-known 'regimentation' effect. A small amount of Type B tissue, i.e., clumps of cells of no fixed pattern which show vacuolation in varying degrees to give the so-called microcystic degeneration, was also present. At no place was there any evidence to suggest malignant change. No nerve-fibres were seen."

One section stained H. and E. is shown (*Fig.* 384).

PROGRESS.—One year later the patient was in excellent health. Wasting of the tibialis anticus was still present

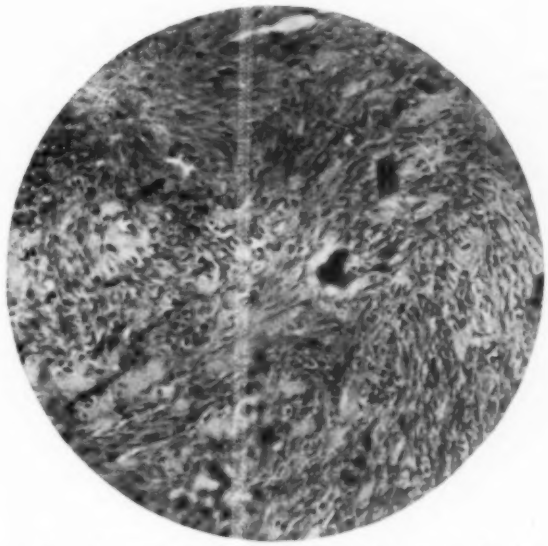

FIG. 384.—Histological appearance of the tumour showing both regimented and loose grouping types of arrangement of its cells.

and dorsiflexion was still done by the extensors digitorum and hallucis longus and peroneus tertius. Inversion was a little weak, but plantar flexion and all the other movements were excellent. He wore the spring to prevent fatigue after a long day's work as a butcher.

DISCUSSION

The diagnosis of such tumours is seldom made in the absence of disturbance of nerve function, especially in the less accessible and infrequent situations. In this case the diagnosis was not considered, as the nervous manifestations did not appear until after an operation, which revealed a lesion which appeared inflammatory because of its acute onset, the presence of congestion, œdema, and of adhesions accompanied by a vague sense of fluctuation, and of a tumour of large size (it is one of the larger ones) causing disturbance of function of neighbouring organs—all calculated to lead one off the trail. The acute onset must have been due to some marked degenerative process, hæmorrhagic or thrombotic, which subsided, as its signs diminished enormously between operations, and there was little evidence of recent acute hæmorrhage or necrosis in the histology six months later, apart from the slower microcystic degeneration. Further painful sensations may have been added by abnormal function in the neighbouring organs, e.g., colon (diarrhœa) and bladder (retention), or by the factors which caused this dysfunction—either direct irritation from the exudates accompanying the degenerative process which caused the adhesions, or pressure on the sympathetic and parasympathetic nerves in the pelvis.

Some degree of fluctuation is quite a well-known phenomenon in such apparently solid tumours, possibly imparted by the numerous tiny cystic spaces.

As regards surgical removal, in this case no nerve of any kind was felt as the tumour was shelled out, therefore it must have arisen from such a small, unimportant branch that even its presence was unsuspected. No further nerve impairment followed, extirpation confirming this. Nerve-fibres are never found inside the capsule, but stretch over it, in this type of tumour. Stout is of the opinion that if the tumour involves an important trunk, it should be preserved even at the risk of not removing all the tumour, as, in contrast to neurofibromata, the neurilemmomata for practical purposes never become malignant. If a major nerve is damaged it should be repaired so that dysfunction passes off in months or years. In our present case the one muscle has wasted despite intensive physiotherapy, so that even in the presence of an intact nerve, function is unlikely to recover completely. It is a very minor disability even now. The prognosis seems excellent otherwise.

SUMMARY

A neurilemmoma of the pelvis has been described, its various unusual features detailed and discussed, especially its rarity, its acute onset, and its involvement of both spinal and autonomic nerves.

I have to thank Mr. R. L. Benison for permission to report this case; Dr. Hewspear for the pathological report, and Mr. Paton for the photographs.

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In Memoriam

A. RENDLE SHORT

M.D., B.S., B.Sc., F.R.C.S.

BY the sudden death of Professor Rendle Short, Bristol, and indeed the surgical profession generally, have lost one of their outstanding personalities. Rendle Short was born in Bristol on Jan. 6, 1880, and received his education at Bristol Grammar School and University College, which he entered in 1897. He took a London B.Sc. degree, with First Class Honours in Geology, before proceeding to the clinical part of the medical course. His undergraduate period was crowded with distinctions —Gold Medals in Physiology, Materia Medica, and Medicine, and First Class Honours in Anatomy and Obstetrics; in fact, the only subject in which he did not claim a distinction was surgery, which he was to make his life's work. He took his medical degree in 1903, followed this with M.D. with Gold Medal in 1904, and was admitted to the F.R.C.S. in 1908.

He was appointed to the Staff of the Bristol Royal Infirmary in 1913, became full surgeon in 1922, and retired in 1946 when he was appointed Honorary Consulting Surgeon. In Bristol University he was demonstrator and subsequently Lecturer in Physiology from 1910 to 1928, clinical Lecturer in Surgery from 1914 to 1932, and Professor from then until his retirement in 1946. From then until 1953 he acted as Medical Officer to the cancer service, at first to the Counties of Somerset, Gloucester, and Wiltshire, and then to the South-Western Regional Hospital Board. Perhaps his greatest service to British Surgery was his advocacy of the physiological approach, for he was one of the pioneers of the application of modern physiology to the practice of surgery, and his book *The New Physiology in Surgical and General Practice*, first published in 1922, went through five editions. A service to the Medical Profession as a whole was the part he played in the editorship of the *Medical Annual*.

His knowledge in many directions was astounding. Though very few people realized it, one of his greatest interests was history, and he read widely in the subject up to the end. In addition, of course, he had a profound knowledge of geology and always carried a small hammer in his pocket to add to the collection of specimens in his study. It was characteristic of him that during his last visit to Southmead Hospital he was found by his house surgeon at the bottom of a deep trench, which was being sunk to take a new drainage system, hammering away at some fossils which he had noticed while on the way up to the theatre. He did not take any interest in ornithology until he was about 25, when out for a walk with a companion he found that his friend could recognize the song of a number of birds. He immediately commenced observing birds and very soon gained a vast knowledge of the subject.

No fact which he wished to remember seemed to be lost, and if he did not know the answer to a question he could give chapter and verse where it could be found.

He was experienced in natural history in all its branches and wrote for the lay public on ancient

linguistics, biology, evolution, and archæology, though he would rarely speak of them.

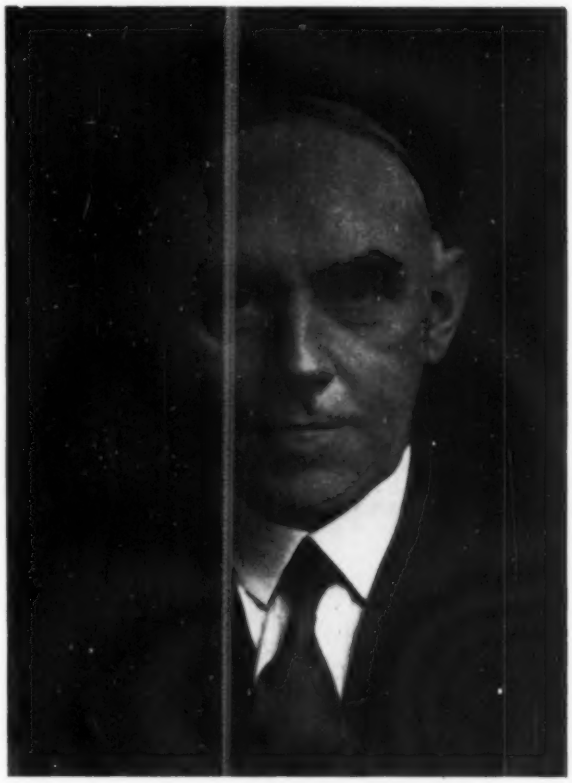
He gave a Hunterian lecture just before the 1914–18 War on surgical shock. It was a most propitious time for the subject, though he did not know it. Consequently he was given a roving commission across the Western Front to investigate the subject further, and a good deal of his roving was done on a bicycle. He often told his students about his first use of blood transfusion at Poperinghe and the lucky outcome in view of the fact that he did not know anything about the blood groups at the time. He had a great sense of the dramatic and his descriptions of the tented wards of a C.C.S. set aside for the treatment of the severely shocked were most vivid.

Undoubtedly his greatest asset to the Bristol Medical School was his undergraduate teaching, at which he was a master. The older of his past students remember him first in the Department of Physiology, where his outstanding lectures on the Central Nervous System demonstrated that a most difficult subject could be presented in a manner which all could understand and retain. His clinical teaching was of the same order—clear, incisive, dogmatic; with complete absorption in the task of the moment, he would pick out the salient points in each case and drive them home. He was best on the well-known conditions of every day and would bring in some of his funny anecdotes whenever possible, starting always with: " I well remember ". Among his most attractive traits was his sense of humour, which crept into almost all his teaching, and there must be many who will never forget his apposite similes. " The tonsils always remind me of a pair of ' chuckers out ' at a place of popular entertainment. They stand, one on each side of the door, to deal with any rowdy customers that may come along. It is easy to understand that if the ' chuckers out ' be old and stout they get the worst of the encounter. " This, with a twinkle in his eye, for it is doubtful if he ever went to a place of popular entertainment and certainly not one of the variety which necessitated the employment of " chuckers out ".

On Mondays at noon he gathered the final year students around him and by question and answer and oft-quoted aphorisms would awaken interest in the most lethargic minds. He knew all the students by name and could usually give a fair outline of their background and followed them keenly throughout their careers. When he knew that one of them was in pecuniary difficulties he would, if they had any aptitude, ask them out to assist in private for which he gave them immediately afterwards the sum of one guinea, usually paid as one pound note, one sixpence, and six coppers.

He was very fortunate in having a robust constitution and travelled over the West Country to give an opinion or to operate in farm houses, cottages, or mansions. He used to travel to hospital and to consultations in the Bristol area on a bicycle in all weathers, and it was not until 1930 that he invested in a car. When called at night to do some emergency operation, the hospital used to send a cab, but he preferred to cycle because, as he would say, he was so poor in his early days that he could not afford to tip the cabbie. He revelled in hard work—night or day.

In 1916 in Bristol he recorded that he did 54 operations in three days—at that time he was doing nearly all the night emergencies at the Bristol Royal



A. RENDLE SHORT
(1880–1953)

Infirmary and had charge of half the surgical Wards and Out-patients.

In manner he was abrupt, and sometimes there appeared a certain awkwardness and aloofness in his approach. The fact of the matter was that he was an extremely shy person and found it difficult to unbend. It was this shyness and the determination to put every second to the greatest possible use that made him so curt at times. He did not suffer fools gladly, but his great humility would often cause him to go back and apologize if he thought he had been too censorious.

No estimation of his personality and character would be adequate without mention of his deep and simple Christian faith based on a vast knowledge of, and trust in, the Bible. For this he was known almost all over the country, as he used to speak widely on the subject. His last book, *Wonderfully Made*, written for the intelligent laity, is a definite attempt to point to the hand of God in the fashioning of all living things and particularly in directing the complex mechanisms of the human body, notably

those which have not yet been explained on purely scientific grounds. He was completely unashamed of his faith, though his shyness prevented him from discussing it in ordinary conversation.

His outstanding characteristics, however, were his abounding love for his fellow men, which he was too reserved to express in words, and his steadfast integrity throughout his life.

VICTOR BONNEY

B.Sc., M.D., M.S., M.R.C.P., F.R.C.S., F.R.C.O.G., F.R.A.C.S.

IT is appropriate for this JOURNAL to pay tribute to the memory of Victor Bonney for he was first and foremost a surgeon and his death has left a gap in the world of surgery.

He entered Middlesex Hospital in 1891; as a student it is said that he was not a hard worker, but

VICTOR BONNEY
(1873–1953)

he had a most retentive memory and this made him a good examinee and later a fine teacher. He wanted to be a physician, but his steps were directed towards gynæcology by John Bland-Sutton, and he went to Chelsea as Resident Officer. In most hospitals gynæcology was regarded as coming within the province of the physician, and a general surgeon was called in if laparotomy was necessary. At Chelsea the reverse opinion held, and the gynæcologists did their own operating; their range covered all the pelvic and abdominal contents and the mammary regions. When Bonney was appointed to the staff there in 1901 he had the title changed from gynæcological physician to gynæcological surgeon, a procedure which he later repeated at Middlesex. He always felt that gynæcology should be a part of general surgery but that it should be perfected and maintained in cohesion with the other specialties in order to perfect surgery as a whole. His own urge to perfection was mirrored in his Hunterian Oration of 1943. Amongst the many honours he received none gave him greater pleasure than his election as Honorary Fellow of the Association of Surgeons.

He was the first gynæcologist to be elected to the Council of the Royal College of Surgeons of England, and it was largely through his advocacy that specialist surgeons were co-opted to serve on the College Council.

During the 1914–18 war he worked at the Middlesex convalescent home at Clacton, and whilst there he successfully ligated the innominate artery. His skill as an operator was superb, but was equalled by his care for his patients afterwards. His kindliness was apparent to all who came in contact with him and his enthusiasm was infectious. He maintained his interest in surgery to the end, and only a few months before his death he paid a visit to the operating theatre to see something which was new to him, in order to keep abreast of the times.

That he was ill at the time arranged for the dinner to celebrate his eightieth birthday was a source of regret to his colleagues as much as to him, but he has left a record of the speech he was going to make on that occasion. It gives a true estimate of the man. To use his own quotation from St. Paul he "continued in the faith".

EXPERIMENTAL SURGERY

OBSERVATIONS ON THE HEALING OF EXPERIMENTAL GASTRIC ULCERS IN SMALL LABORATORY ANIMALS

By A. WYNN WILLIAMS

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In a consideration of the pathogenesis of chronic peptic ulcer, Ivy, Grossman, and Bachrach (1951) have aptly written: "It is difficult to conceive, and no one has explained adequately, how a chronic ulcer could arise, like Minerva springing fully armed from the forehead of Jove, without it being initially an acute lesion of the mucosa." A study of the normal and abnormal healing of acute peptic ulcers is clearly of importance. Spontaneous peptic ulceration is rare amongst domestic, laboratory, and wild animals. Hutyra and Marek (1913) refer to the rare occurrence of peptic ulcer in adult cattle, dogs, swine, and horses. Bongert (1912) found gastric ulcers in weaning calves. Fox (1923) reported the occasional occurrence of chronic gastric ulcer in Primates, Carnivora, Primipedia, Insectivora, Ungulata, and Hydroces. Schroeder and Wegeforth (1935) found a high incidence of chronic ulcer in Californian seals.

Peptic ulcers have been reported in dogs (Rosenow 1923; Overgaard, 1934; Keller, 1936) and rats (Singer, 1913), but not in guinea-pigs (Smith and McConkey, 1933) or rabbits (Ophüls, 1906).

Although spontaneous peptic ulcers occur rarely or not at all in laboratory animals, the study of the healing of experimental peptic ulcers in these animals is of considerable biological interest, although, of course, conclusions from such experiments are not necessarily applicable to human beings.

The study of healing of defects in the gastro-intestinal tract has not been as thorough as the subject warrants. Most investigations have been on the dog. No essential difference between the healing of an acute peptic ulcer in man and that described for the dog and cat has been found (Stewart, 1922; Caylor, 1926; Kennedy, 1926).

Experimental peptic ulcers have been produced by many different methods. Excision is probably the commonest of the physical procedures. Acute ulcers have been produced by cauterization with a hot iron in the stomach or duodenum of rabbits by Quincke (1875), Yano (1925), Nakano (1932) and Oda (1936). Cohnheim (1882) also produced acute lesions of the stomach of rabbits by burning. These lesions heal in the same time as ulcers produced by excision. By burning the stomach mucosa under standardized conditions and later fixing the stomach in situ with a constant volume of fixative, it is possible to obtain an accurate assessment of the healing of defects of the gastric mucosa. Such a method is now described. This description is followed by some examples of the application of the method and the results obtained.

METHOD

The animal is given a general anæsthetic and the abdomen opened through a midline incision. The stomach is withdrawn from the abdomen and a small incision made in the anterior wall at right angles to the long axis of the stomach. The posterior wall of the stomach is then applied to the newly-made

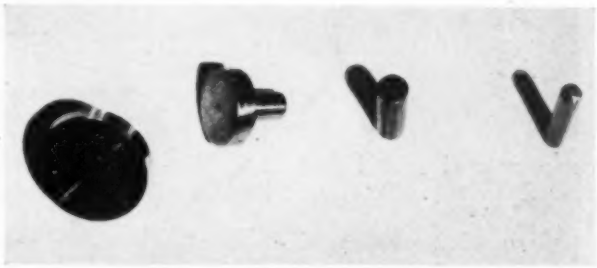


FIG. 385.—Instruments for producing ulcers. (× ¾.)

aperture in the stomach wall by a finger placed behind the stomach. With the finger-tip remaining behind the elected site of trauma on the posterior wall, the wall is cauterized with a stainless steel cylinder. In the experiments, the brands (*Fig.* 385) varied from 1·0 to 18·0 mm. in diameter. They were heated for a constant time, e.g., 20 to 30 seconds, in a small flame of constant temperature. The brand is applied, with slight pressure, to the mucosa until the sensation of heat felt by the underlying finger-pad becomes unbearable. After burning, the stomach wall is returned to its natural position, and the aperture in the stomach wall is then closed with sutures—a purse-string suture is simplest and safest. Finally, the abdominal wall is closed in two layers.

The burn is a well-defined lesion. The whole operation occupies, as a rule, no more than twenty minutes. When it is desired to ascertain the progress of healing at a given time after operation, the animal is killed and its duodenum and œsophagus are tied. Its stomach is then filled with a constant volume of fixative, the amount being appropriate to the size of animal, e.g., 8–10 c.c. for young adult rats, 30–40 c.c. for young adult guinea-pigs, and 60–80 c.c. for young adult rabbits. About one hour after filling the stomach, the latter is removed and immersed in fixative for 24 hours. When fixation is complete, the stomach is opened and the mucosa washed very gently with a small stream of running water. The healing or healed lesion is usually easily identifiable on a relatively flat background.

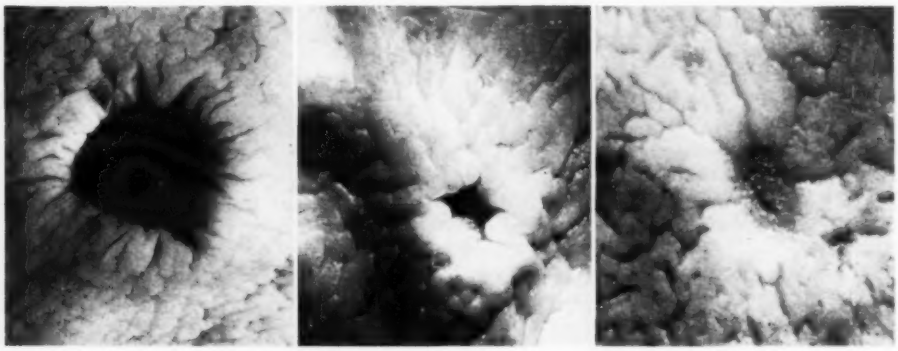
APPLICATION OF METHOD

The method has proved easily applicable to guinea-pigs, rabbits, and rats. Observations were made on the healing time of lesions of the same size in these animals; others were made on the influence of size of lesion on healing time, also on the healing time of multiple lesions. The healing process was studied microscopically. The animals were always on a normal mixed diet and their weights were recorded daily. All the animals used were young adult females.

Size of Lesion and Healing Time.—Using groups of 10 to 30 animals for each experiment, ulcers of various sizes were burnt on the middle of the posterior wall of the body of the stomach, midway between the curvatures.

It was found that ulcers 4 mm. in diameter healed completely in guinea-pigs in 13–17 days and in the same time in rabbits. But in rats healing was not complete for 60 to 40 days. This finding in rats may be related to the site of the lesion, which was close to the main arterial supply of the stomach (Berg, 1947). Smaller lesions in rats (1 mm. diameter) healed completely in 15–20 days.

In guinea-pigs and rabbits lesions 2·5 mm. in diameter healed in the same time as the 4 mm. lesions, while large ulcers (12 mm. diameter) healed completely in 15–30 days. Very large ulcers (18 mm. diameter) healed completely in guinea-pigs and rabbits in 25–40 days.

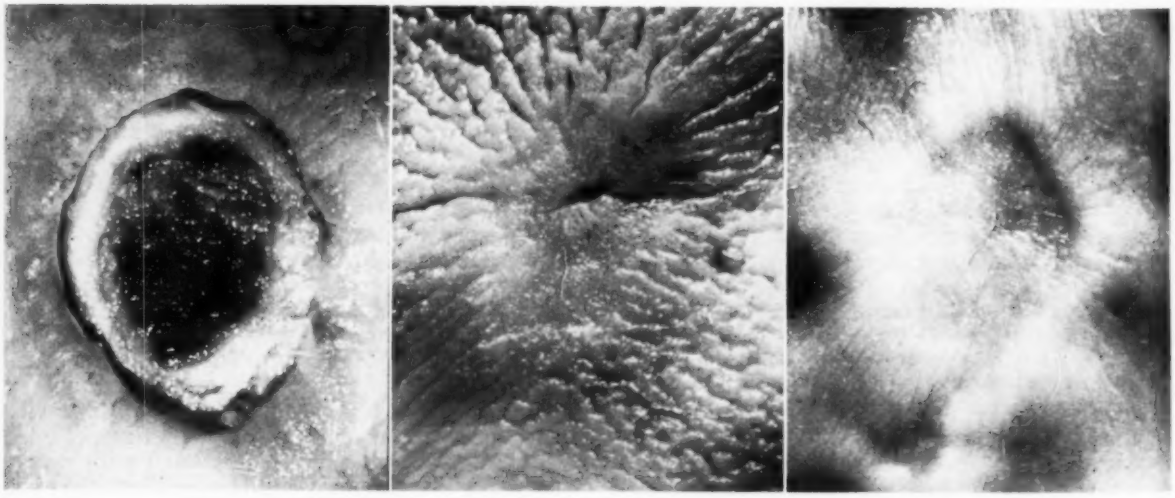


A B C

FIG. 386.—Healing of ulcer initially 4 mm. in diameter in stomach of guinea-pig. A, At 4 days; B, At 8 days; C, At 16 days. (× 4·5.)

The healing of lesions is illustrated in *Figs.* 386–390.

Body-weight and Healing.—The general pattern of the post-operative fall and subsequent rise is similar in the various animals (*Figs.* 391–393); and it is of considerable interest that the intense fibroblastic proliferation begins at the time that the body-weight is rising.



A B C

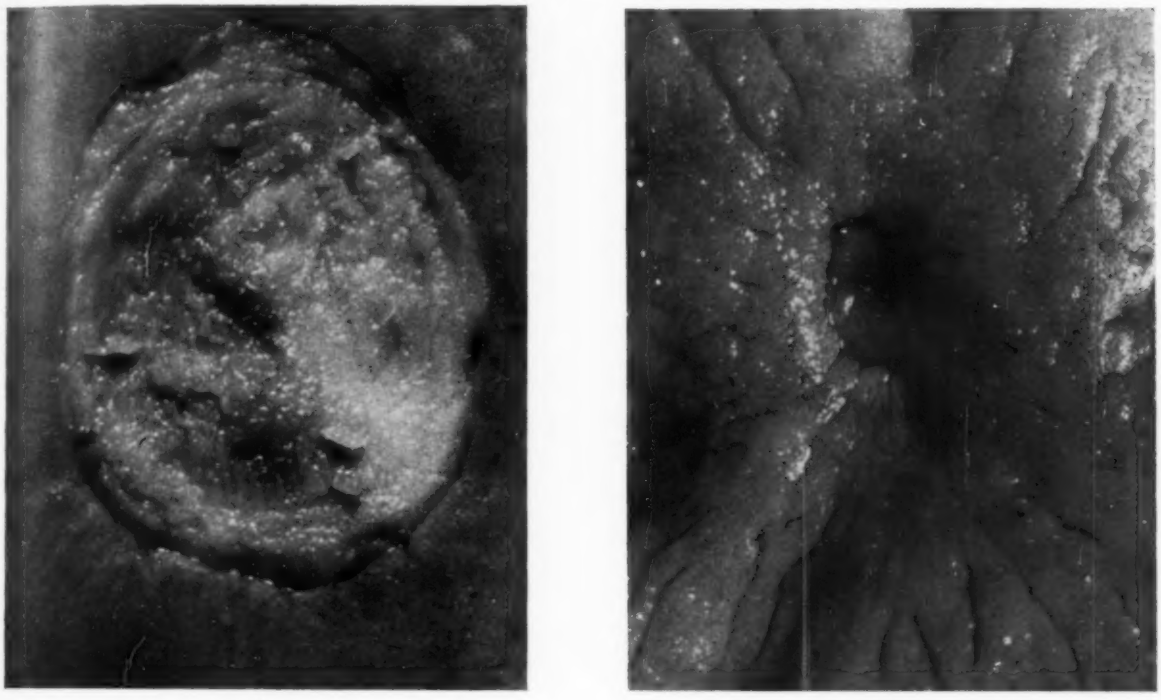
FIG. 387.—Healing of ulcer initially 12 mm. in diameter in stomach of guinea-pig. A, At 6 days; B, At 20 days; C, At 23 days. (× 4·5.)

Site of Lesion and Healing Time.—This relationship was studied in the guinea-pig. Lesions 4 mm. in diameter were burnt in the stomachs of three groups, each containing 12 animals. In the first group the ulcer was at the œsophagogastric junction, in the second it was on the middle of the

lesser curvature; and in the third group it was at the gastroduodenal junction. The animals were killed on the seventeenth day. The lesions at the œsophagogastric junction and on the middle of the lesser curve had healed completely. Eight of the lesions at the gastroduodenal junction had healed completely. The remaining four were almost, but not entirely, healed.

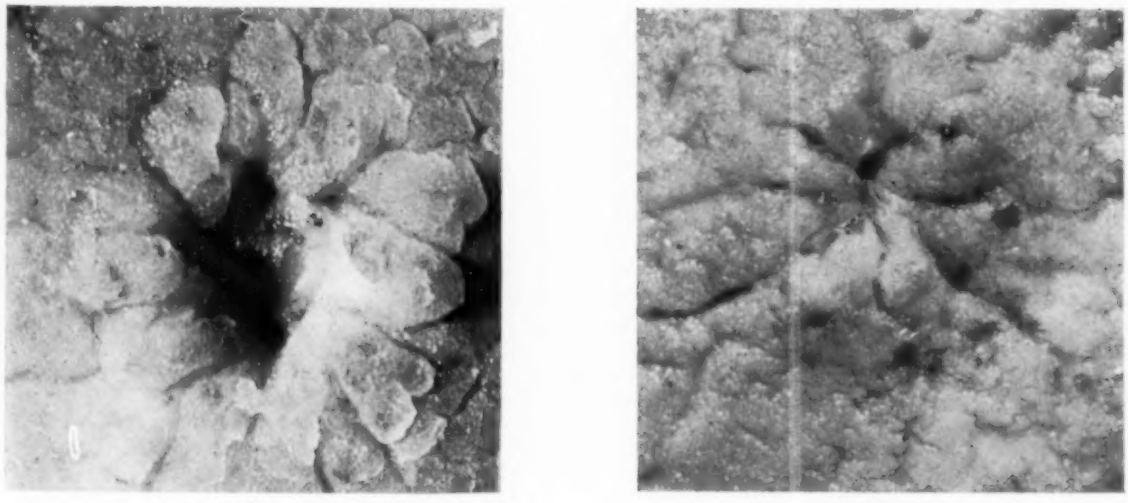
Multiplicity of Lesions.—Using 6 animals for the experiment, it was found that three 4 mm.-diameter ulcers healed as quickly as a single lesion of the same size in the guinea-pig. The same result was obtained in 6 rabbits.

Microscopical Changes.—The pattern of healing is identical in all three animal species. It comprises two components: epithelial regeneration and connective-tissue formation. The former begins within 24 hours of injury, when a delicate film of epithelium, one cell thick, grows over the ulcer floor from the neighbouring gastric pits. At first the cells are flat, later they become cuboidal, and, finally, columnar in shape. It is commonly stated in reports



A B

FIG. 388.—Healing of ulcer initially 18 mm. in diameter in stomach of guinea-pig. A, At 6 days; B, At 27 days. (× 6.)



A B

FIG. 389.—Healing of ulcer initially 4 mm. in diameter in stomach of rabbit. A, At 8 days; B, At 15 days. (× 6.)

and text-books that such epithelium can grow only over a firm, connective-tissue base; but it has been observed many times in these experiments that the cells can and do grow over necrotic debris, providing such debris forms a uniform covering for the floor.

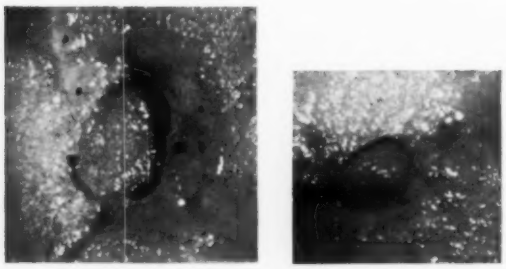


FIG. 390.—Healing of ulcer initially 1 mm. in diameter in stomach of rat. A, At 5 days; B, At 9 days. (× 9.)

This epithelial covering is probably of considerable importance in the healing process because it protects the floor of the ulcer. The initial acute inflammatory response wanes after 48 hours. There is little fibroblastic proliferation until the fourth, fifth, or sixth days, when there is a sudden burst of activity. This proliferation, as would be expected, is most marked in the connective-tissue planes. Several days from the beginning of ulceration, collagen

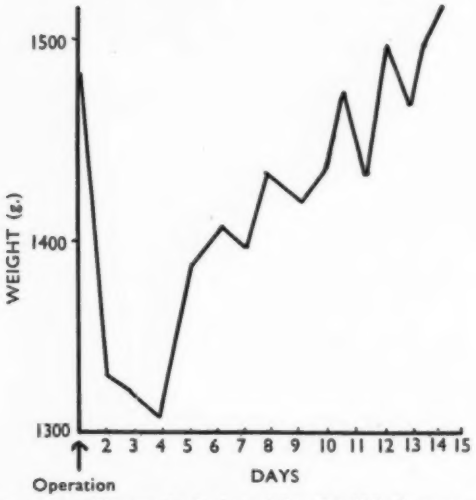


FIG. 392.—Changes in body-weight of rabbit (average of 12 animals) following production of gastric ulcer.

is formed. A strong, fibrous tissue is produced in an orderly manner. At the same time the covering epithelium differentiates into glandular tissue, at first consisting only of a few simple tubules, later of fairly thick mucous membrane which is pyloric in type. No differentiation into chief and parietal cells has been seen. Epithelial proliferation is particularly well marked in the rabbit. In this animal the fully-formed new mucosa appears to the naked eye as a boss of tissue. In all the animals studied the mature, new mucous membrane was well-formed. Occasionally there was a minor degree of cyst formation or polyposis.

Phases of the healing process, as seen microscopically, are illustrated in *Figs.* 394–405.

DISCUSSION

Most studies on wound healing have concerned wounds of the skin. Arey (1936) stated that internal wounds of mucous surfaces differed in no fundamental way from those of external surfaces. The studies described here support that opinion. He noted that epithelization ceased if, after contraction of the wound, the area remaining to be covered exceeded 2·5 cm.

All the epithelial cells of the stomach arise from a mitotic focus located at the bottom of each crypt (Harvey, 1906; Katayama, 1920; Bensley, 1928). Grant (1943) has described migration of epithelial cells to fill gaps in the stomach mucosa one or two hours after the injury of the mucosa of cats by 50

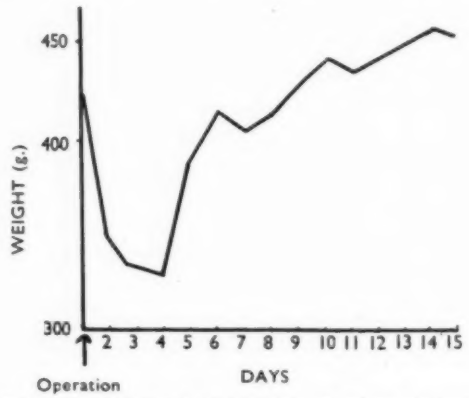


FIG. 391.—Changes in body-weight of guinea-pig (average of 20 animals) following production of gastric ulcer.

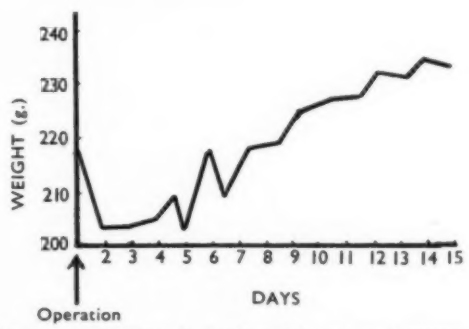


FIG. 393.—Changes in body-weight of rat (average of 15 animals) following production of gastric ulcer.

per cent alcohol or 1 per cent acetic acid; and in the experiments described here cell migration was observed within 24 hours of injury. It is commonly held that these epithelial cells migrate only over healthy granulation tissue and not over necrotic material; but in several instances epithelial cells were seen to have grown over necrotic material which was condensed and continuous.

Griffini and Vassale (1888) investigated the healing of acute ulcers in the dog's stomach. They excised pieces of mucosa and studied the specimens microscopically for up to 55 days after the operation. They noted a decrease in the size of the defect by

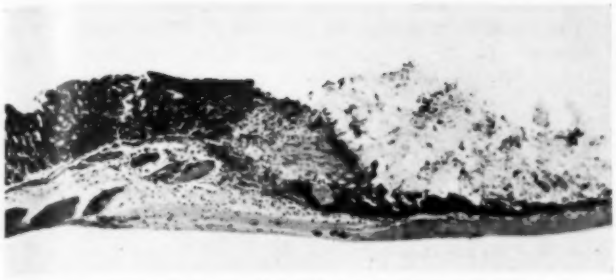


FIG. 394.—Ulcer in guinea-pig stomach on second day. H. and E. (× 35.)

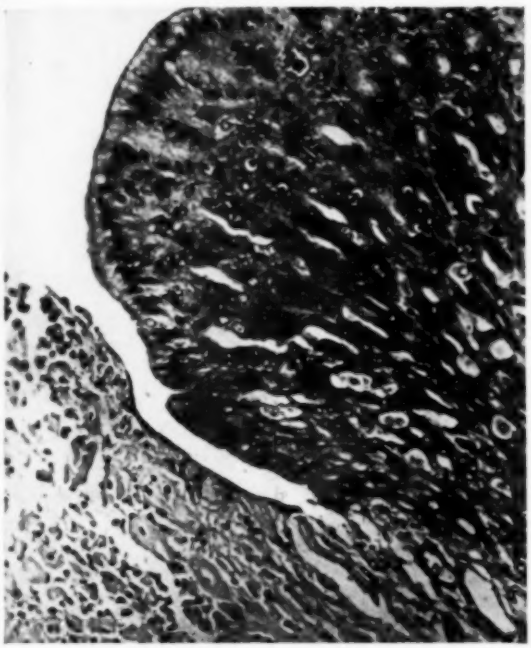


FIG. 395.—Margin of ulcer in guinea-pig stomach on fourth day. The epithelium is spreading over necrotic tissue. H. and E. (× 135.)

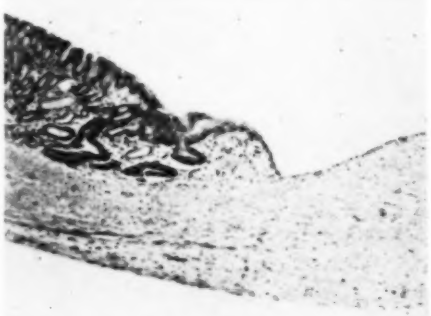


FIG. 396.—Margin of ulcer in guinea-pig stomach on eighth day. H. and E. (× 50.)

FIG. 397.—Healing ulcer in guinea-pig stomach on thirteenth day. The base is entirely covered with epithelium. H. and E. (× 40.)

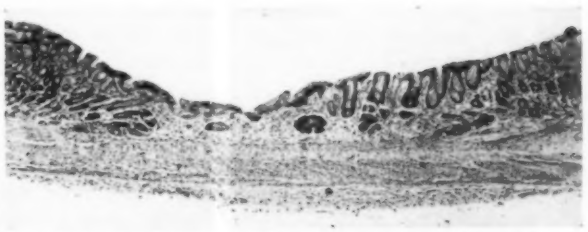


FIG. 398.—Healing ulcer in guinea-pig stomach on fourteenth day. The regenerated epithelium is differentiating into simple tubules. H. and E. (× 40.)

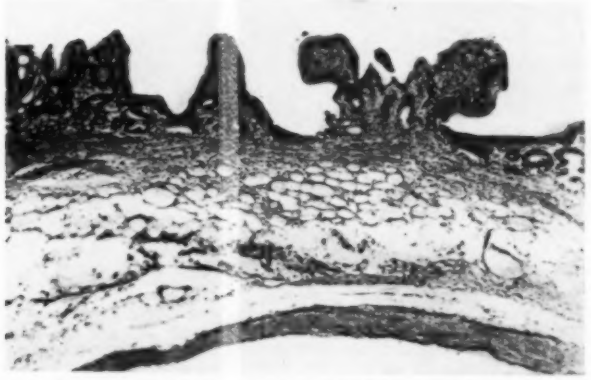


FIG. 399.—Healed ulcer in guinea-pig stomach on fourteenth day. Note the polypoidal development of regenerated epithelium. H. and E. (× 40.)

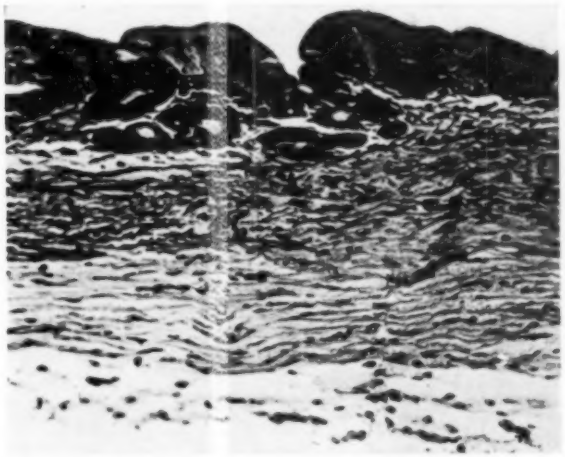


FIG. 400.—Healing ulcer in guinea-pig stomach on fifteenth day. H. and E. (× 112.)

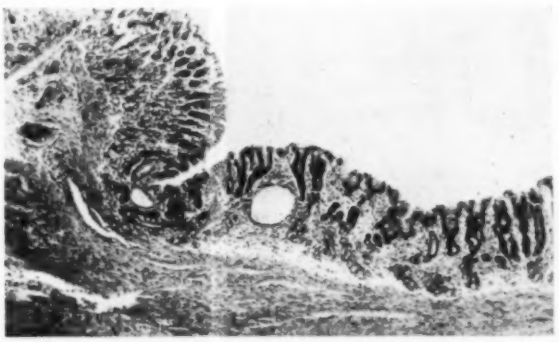


FIG. 401.—Healed ulcer in guinea-pig stomach on twenty-third day. Note the development of glands of pyloric type. H. and E. (× 40.)

muscular contraction immediately after the operation, then the formation of granulation tissue and an epithelial covering. From the latter, new glands were

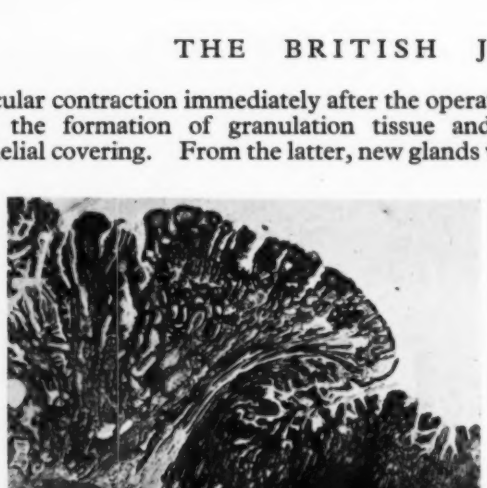


FIG. 402.—Healed ulcer in stomach of rabbit on twenty-fifth day. The reconstituted mucosa (pyloric in type) on the left is greater in thickness than the normal mucosa (body type) on the right. H. and E. (× 30.)

formed. Lim (1922) noted the similarity of the reparative process to that of embryonic development. The similarity is also illustrated in *Figs.* 404, 405. Bolton (1910) described similar findings in the healing of acute gastric ulcers produced in cats by the injection of 'gastrotoxic' serum; and Ivy (1920) made similar observations in dogs on the healing of acute

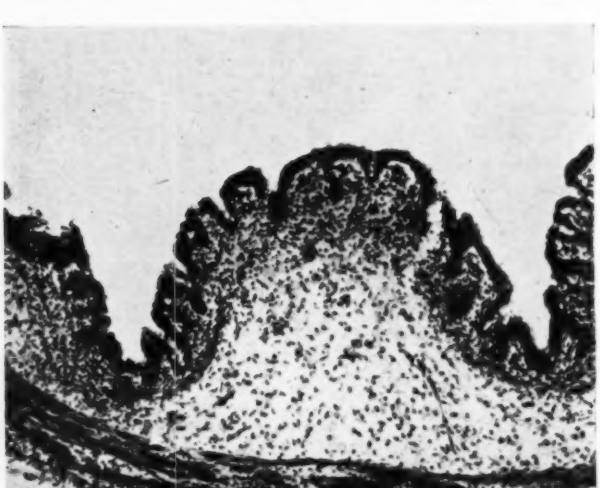


FIG. 404.—Section of human embryo stomach (12 weeks). H. and E. (× 40.)

gastric and duodenal ulcers, produced by excision or by the injection of silver nitrate.

In these present studies healing occurred completely, even in the ulcers 18 mm. in diamter. Wounds 4 mm. in diameter healed in the same time (13–17 days) as wounds 2·5 mm. in diameter in rabbits and guinea-pigs. In the same animal species wounds 12 mm. took longer to heal (22–26 days), and wounds 18 mm. in diameter took 35–40 days. In the larger wounds, however, healing was

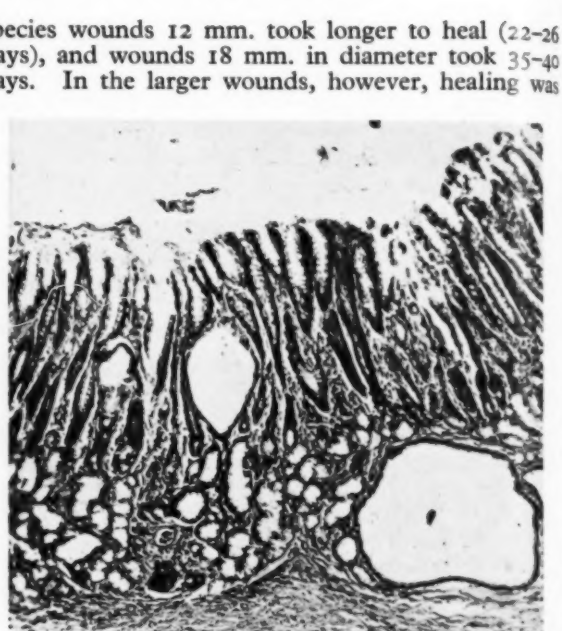


FIG. 403.—Healed ulcer in stomach of rat on sixtieth day. The reconstituted mucosa is pyloric in type. H. and E. (× 75.)

nearly complete 7–10 days before the deficiency was closed. Young, Fisher, and Young (1941) confirmed that large wounds of the skin in rabbits heal at a

FIG. 405.—Section of human embryo stomach (25 weeks). H. and E. (× 40.)

greater rate than smaller ones. In the stomach of animals complete healing may be retarded by muscular movement and the passage of food, both of which would tend to abrade delicate growing epithelium.

The slow rate of healing in the 4-mm. ulcers on the posterior wall of the stomach of the rat is

unexplained but may be due to the proximity of the burn to the main arterial supply of the stomach.

After 4–6 days, intense fibroblastic proliferation begins in the wounds. In 1932, Cuthbertson described the brisk catabolic phase following operation which, after a few days, changes rather abruptly to a less intense but longer period of anabolism. Moore (1953) believes the early catabolism to be due to hormones, and points out that its cessation is marked by features—diuresis and transient eosinophilia—they are observed on withdrawal of cortisone administration. It is at this stage, too, that wound healing begins in earnest.

Most studies on the rate of healing of acute ulcers in experimental animals have been on the dog. Dragstedt (1917) reported that acute ulcers 1·0–1·5 cm. in diameter in the mucosa of a Pavlov pouch in dogs healed in 5–15 days, while ulcers of similar size in the duodenum healed in 20 days. Ivy (1920) observed that, in dogs, ulcers of this size healed in 9–13 days in the body mucosa, in 12–18 days in the pyloric mucosa, and in 16 to 24 days in the duodenum. Morton (1927) observed in the dog that defects made in the body, pyloric, and duodenal mucosa, 2 cm. in diameter, healed in 2 weeks; and he stated that, under adverse conditions, ulcers on the lesser curve healed more slowly than those on the greater curve.

Yano (1925), using rabbits, found that ulcers made with a hot iron healed more rapidly when made in the body of the stomach than along the lesser curve. Walker (1931), however, found that excision ulcers in the rabbit stomach healed more rapidly in the pylorus than in the body.

McIlroy (1927) observed in the cat that defects made in the pyloric mucosa healed more slowly than ulcers made elsewhere in the stomach. Bolton (1913) found no difference in the rate of healing of ulcers produced in different parts of the stomach.

The largest evidence suggests a gradient of healing potential inversely proportional to the gradient of susceptibility to acid injury. In the present studies there was some delay of healing at the pylorus in guinea-pigs.

In man, the healing of acute gastric or intestinal ulcers has been studied in surgical and autopsy specimens (Bolton, 1913; Stewart, 1922; Caylor, 1926; Hauser-Erlanger, 1926; Tanner, 1944). No essential difference between the healing of an acute ulcer in man and that found in animals (dog and cat) has been found (Caylor, 1926; Hurst and Stewart, 1929). Moreover, it appears that in man and animals, under favourable conditions, acute and chronic ulcers heal at almost the same rate (Ivy and others, 1951). Under ordinary conditions acute ulcers of the stomach and duodenum of experimental animals heal rapidly. In man, too, it appears that acute ulcers 1·0–1·5 cm. in diameter usually heal in 3–4 weeks; and the best available evidence indicates that chronic gastric or duodenal ulcer heals in 14–100 days (average 40 days) under strict medical management.

SUMMARY

1. A standardized method for studying the healing of acute gastric ulcers in small laboratory animals is described.

2. Ulcers 4 mm. in diameter healed completely in guinea-pigs and rabbits in 13–17 days and in 40–60 days in rats. Ulcers 2·5 mm. in diameter healed in the same time as the 4-mm. lesions in guinea-pigs and rabbits, while ulcers 12 mm. and 18 mm. in diameter healed completely in 15–30 and 25–40 days respectively in these animals.

3. In the guinea-pig, three ulcers, each 4 mm. in diameter, healed as quickly as one; and an ulcer of this size healed as quickly at the œsophagogastric junction as in the body of the stomach, but at the pyloroduodenal junction there was some slight delay in healing.

4. The microscopical features of the healing process are briefly described.

5. Reference is made to the findings of other investigators.

I am grateful to Professors W. D. Newcomb and J. S. Young for encouragement and suggestions, and to all who have given technical assistance.

APPENDIX

EXPERIMENTAL DATA

Key :—
C denotes *complete healing*.
N denotes nearly-complete healing (defect pin-head size or less).
U denotes *unhealed lesion* (defect larger than pin-head size.)

LESIONS INITIALLY 18 MM. IN DIAMETER

ANIMAL	DAY 40	DAY 30	DAY 25
Guinea-pig	5 C	1 C 4 N	3 N 2 U
Rabbit	5 C	4 C 1 N	4 N 1 U

LESIONS INITIALLY 12 MM. IN DIAMETER

ANIMAL	DAY 30	DAY 25	DAY 20	DAY 15	DAY 12
Guinea-pig	5 C	4 C 1 N	1 C 3 N 1 U	2 N 3 U	5 U
Rabbit	5 C	4 C 1 N	2 C 2 N 1 U	3 N 2 U	5 U

LESIONS INITIALLY 4 MM. IN DIAMETER

ANIMAL	DAY 20	DAY 18	DAY 17	DAY 16	DAY 15	DAY 14	DAY 13	DAY 12
Guinea-pig	—	5 C	5 C	4 C 1 N	4 C 1 N	3 C 2 N	1 C 4 N	5 N
Rabbit	—	—	5 C	4 C 1 N	3 C 2 N	2 C 3 N	1 C 4 N	3 N 2 U

LESIONS INITIALLY 4 MM. IN DIAMETER (*continued*)

ANIMAL	DAY 60	DAY 50	DAY 40	DAY 30	DAY 20
Rat	5 C	2 C 1 N 2 U	2 N 3 U	1 N 4 U	5 U

LESIONS INITIALLY 1 MM. IN DIAMETER

ANIMAL	DAY 20	DAY 18	DAY 15
Rat	5 C	3 C 2 N	5 N

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SHORT NOTES OF RARE OR OBSCURE CASES

ACUTE PERFORATION OF A DUODENAL DIVERTICULUM WITH PERITONITIS

BY GORDON FERGUSON, HASTINGS

DIVERTICULA of the duodenum, though of no great rarity, are not usually associated with symptoms and the *clinique* is, generally, that of their complications rather than of the sacs themselves. The incidence of duodenal diverticula—excluding ulcer diverticula of the first part of the duodenum—has been variously estimated between 22 per cent and 1·4 per cent. Maingot (1948) considers that 5 per cent can be regarded as a fair assessment of the incidence of these pouches for persons aged fifty and over.

Though diverticulosis of the duodenum is relatively common, changes in the pouches themselves or in neighbouring organs secondary to their presence are quite rare. The comparative immunity of these sacs from complications is probably due to (1) Sterility of the duodenal contents; (2) Retroperitoneal position which may allow easy distension; (3) Dependent position of their opening into the bowel; (4) In some cases the opening of the sac is guarded by valvulæ conniventes which may hamper filling; (5) These pouches are well supported by surrounding structures and cannot, therefore, sag easily and cause kinking at the ostium.

The fact of the filling of these pouches with duodenal contents is of importance in determining the onset of complications. The sacs have no muscle coat and cannot forcibly eject any chyme which enters, stasis results, infection follows, and various complications may eventuate. A case of acute perforation is reported here.

Acute perforation of a duodenal diverticulum with generalized peritonitis is a rare complication. It was first mentioned, seemingly, by Wilkie in 1913, but his case was of perforation of an ulcer diverticulum of the first part of the duodenum; such pouches are not nowadays classified as duodenal diverticula. In 1923 Huddy removed a gangrenous diverticulum and in 1926 Monsarrat reported the first case of perforation with generalized peritonitis of a true diverticulum of the second part of the duodenum.

A successful operation was performed. Since these reports no further cases have appeared in the British literature though cases have been reported in American and Scandinavian journals.

CASE REPORT

The patient was a lady aged 58 years who had had a ventrofixation of the uterus in 1925 and who said that she had had treatment for 'blood-pressure'. The history of her illness revealed that five hours earlier she had been seized with sudden severe pain which was epigastric at onset but had by now become generalized. The pain was constant but with exacerbations of greater severity, and in attempting to find relief she had rolled about in the bed. She had vomited once, four hours after the onset of pain, and had retched frequently since vomiting. She had recognized in the vomitus some food which she had taken at midday—it was now 11 p.m.—and there was no blood. Her bowel action was regular: she had had no motion since the onset of pain but had passed flatus. She had not lost weight and had no urinary symptoms. There was no history of indigestion of any kind; appetite was good for all kinds of foodstuffs and there had been no pain after her meals. Findings on physical examination were: T. 97°, P. 100, R. 40, B.P. 110/65 (despite history of 'blood-pressure'); her skin was cold and clammy and she appeared cyanosed; tongue clean and moist; mitral presystolic murmur; there was a subumbilical midline scar, at the lower end of which was a tender firm protrusion which the patient denied ever having seen before the onset of the pain, it was not tympanitic and did not reduce; there was generalized tenderness with two points of greater intensity—the above-mentioned protrusion and in the epigastrium; there was generalized guarding of the abdominal muscles, though at no point was there board-like rigidity; the abdomen was tympanitic; there was no distension and intestinal sounds, not increased in any way, were heard on auscultation; the abdomen moved only slightly on respiration, which was shallow and gasping in character; the phenomenon of rebound tenderness was not conclusively elicited; on rectal examination no mass was felt and there was no point of tenderness; the hernial orifices were normal. A differential diagnosis as follows was arrived at: (1) Perforated peptic ulcer; (2) Acute pancreatitis, (3) Mesenteric thrombosis or embolism.

AT OPERATION.—Laparotomy was performed through a right paramedian incision. On opening the peritoneum there was no escape of gas such as frequently confirms a diagnosis of perforated ulcer. Boland (1939) and Monsarrat (1926) both noted this absence. There was a large quantity of free fluid, stained by intestinal contents, in the peritoneal cavity. The stomach and the first part of the duodenum were normal. An œdematous hæmorrhagic mass was found lying in the concavity of the duodenum on the head of the pancreas. This was found to be a duodenal diverticulum arising by a narrow neck from the second part of the duodenum in close proximity to the ampulla of Vater; it was about 1½ in. in length and consisted of mucous membrane with a thin fibrous covering. The diverticulum was œdematous and friable and had perforated close to its blind extremity. The pouch was dissected from the pancreas without difficulty and its neck identified; it was here ligated and excised. An attempt to invaginate the stump was not entirely successful owing to the friable œdematous state of the tissues; it was covered by approximating the remains of the adventitial coat over it, and some adjacent omentum was utilized for the same purpose. The tender swelling in the lower abdominal scar was found to be the uterus. The wound was closed in layers and drainage was provided through a stab wound in the right flank. The patient made a good recovery and when subsequently seen in the out-patients department was fit and well, had a good appetite, and was gaining weight.

DISCUSSION

The difficulties which arise in treating perforation of a duodenal diverticulum will be realized from the fact that in the cases reported by Boland (1939) and Beaver (1938) the diverticulum was not identified at operation. In the case here reported the pouch was lying in front of the head of the pancreas and thus easily identifiable, but this fortunate state of affairs is unlikely to be often repeated. One must, however, consider the possibility of perforation of such a diverticulum if faced with the problem of an unexplained peritonitis when at operation a diagnosis of, perhaps, perforated duodenal ulcer has proved to be incorrect.

In summarizing his case Boland writes: "I would suggest that if the signs and symptoms point to a perforated ulcer and the intraperitoneal pathology reveals nothing more than an exudate along the anterior duodenal wall a small incision may be made in the posterior peritoneum and a search begun for a diverticulum." It is worthy of note that both Huddy and Monsarrat were guided to their diverticula by an œdematous and infiltrated appearance on the right side of the second part of the duodenum.

SUMMARY

1. A case of acute perforation of a duodenal diverticulum is reported. This is a rare condition which may be difficult to detect even at laparotomy.

2. Acute perforation of a duodenal diverticulum cannot be diagnosed pre-operatively; if at emergency laparotomy for acute peritonitis the diagnosis as to cause is found to be incorrect, and no other obvious lesion is present, a search should be made for a duodenal diverticulum.

3. Œdema and exudate to the right of the second part of the duodenum may be a guide to the correct diagnosis.

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CARCINOMA OF THE TRACHEA WITH COEXISTING FIBROMA OF THE PLEURA

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NEOPLASMS of the trachea are sufficiently uncommon to warrant recording the following case.

CASE REPORT

HISTORY.—A male aged 68, a messenger, was first seen in May, 1951, complaining of intermittent blood-stained sputum for some months, and a chronic cough worse in the winter. He had a harsh inspiratory stridor. Radiographic examination of the chest revealed a sharply defined opacity in the left upper zone, with some areas of calcification. This was rounded in outline, with the

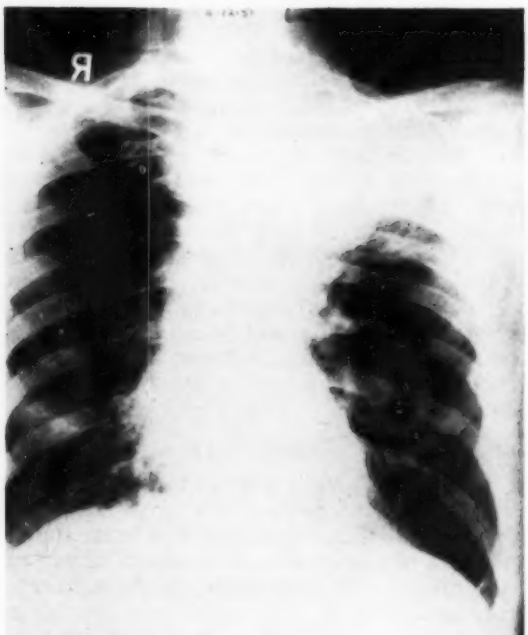

FIG. 406.—Radiograph of chest. December, 1951.

convexity downwards; there was slight displacement of the mediastinum to the right. The radiologist considered the appearances were those of an extrapulmonary rather than those of an intrapulmonary neoplasm. Bronchoscopy showed a nodular swelling of the left lateral wall of the trachea; the remainder of the tracheobronchial tree was normal. Biopsy of this swelling showed a squamous-cell carcinoma.

A clinical diagnosis of malignant teratoma of the mediastinum with invasion of the trachea was made. More complete diagnosis was not possible without thoracotomy, which was considered inadvisable in view of the patient's condition. It was decided to irradiate the mass to relieve tracheal compression. A total dose of 3000 r was given during July and August. When seen a month later there was no change in the clinical state or in the size of the opacity in the left upper zone.

Three months later, in November, the patient was admitted to hospital with a history of having been at work till a week previously, cough with some hæmoptysis for six months, much worse for the last week, and a feeling of weakness for six weeks. He stated that eating made him breathless. On examination the trachea was central, there was a marked inspiratory stridor, there was dullness to percussion over the upper part of the left chest, and the breath-sounds were bronchial over the left lower lobe.

His condition improved during the first few days in hospital but the inspiratory stridor and hæmoptysis continued. Following radiographic examination of the chest and thoracic inlet on Dec. 4 and 5, Dr. Batten reported: "There is a dense mass occupying the left upper zone with areas of calcification in it. This is unchanged since July-August, 1951, and was thought to be extrapulmonary when previously examined and from our findings. There is also evidence of pressure on the trachea by this mass, and displacement to the right and forwards, which seems to have increased a little between the two investigation periods. Except for a very small amount of fluid at the right base, radiological examination

FIG. 407.—Photomicrograph of carcinoma of trachea. (× 5.)

showed no other abnormality; but no special studies of the trachea *per se* were made." (*Fig.* 406.) Endoscopic examination was not considered justifiable in his present state. Bronchopneumonia supervened and the patient died.

AUTOPSY FINDINGS.—The body was that of a somewhat wasted elderly male of average build.

Thorax: A large firm rubbery mass oval in shape and 10 cm. in greatest diameter was present in the inlet of the left thoracic cavity. This was attached to the pleura overlying the arch of the aorta and the vertebral column, but was easily dissected from both. The cut surface showed a whorled appearance, and was yellowish in colour, with a few small calcified areas towards its centre.

The larynx and œsophagus were normal. The tracheal mucosa between the second and eighth rings, a distance of about 3 cm., was thickened, nodular, and ulcerated, and occluded about half its lumen, more particularly on the anterior and left sides. The thyroid was dissected away from the trachea, exposing an infiltrating neoplasm which had invaded the whole thickness of the tracheal wall, the paratracheal tissues, and also the capsule of the thyroid, but the gland itself was free from macroscopical invasion. Enlarged lymph-nodes in the right cervical group showed secondary carcinoma. No distant metastases were found. The lungs were congested and showed chronic bronchitis; the greater part of the left upper lobe was collapsed. Bronchopneumonia was present in both lower lobes. The heart weighed 305 g. and showed normal appearances, the coronary arteries were calcified, the arch of the aorta was distorted by the tumour in the left thorax but was otherwise normal. The

only other abnormality in the body was a mucocele of the appendix.

HISTOLOGY.—The trachea was the seat of a poorly differentiated squamous carcinoma which had invaded the paratracheal connective tissues, leaving the cartilages intact (*Fig.* 407). Microscopically two secondary carcinomata were present in the thyroid, which was otherwise normal. Secondary squamous carcinoma had replaced the greater part of the lymph-nodes from the right cervical group. The mass in the left thoracic cavity showed a partly necrotic and calcified fibroma.

DISCUSSION

Although carcinoma of the trachea was considered as a possibility when the patient was first seen, the prominent benign tumour in the thorax was misleading and prevented the diagnosis from being made with certainty. Had the correct diagnosis been made then it is doubtful whether the patient would have survived excision of the trachea.

The increased discovery of tracheal neoplasms is almost certainly due to better diagnostic facilities, especially the more frequent use of endoscopic examination of the respiratory tract, but the condition does not seem to have increased anything like as much as bronchial carcinoma. Ellman and Whittaker (1947) found a total of 477 recorded cases of neoplasm of the trachea in the literature between 1898 and 1945 and added 1 case of their own. Between 1945 and 1951 75 additional cases of tracheal neoplasms have been found in the British, American, and French literature.

The relative immunity of the trachea to carcinoma as compared with the bronchus may be due to the absence of stagnating secretions with less opportunity for squamous metaplasia to take place (Penta, 1947). Not all workers agree that metaplasia necessarily precedes malignant change; many consider it to be the result of malignant change overtaking the pluripotential basal cells of the mucosa (McDonald, 1946; Guttman, Simon, and Nitka, 1950).

Many writers (e.g., Ide and Cahn, 1948; Ellman and Whittaker, 1947) state that the lower third of the trachea, in its posterior part, is most commonly affected by malignant neoplasms. In the papers consulted, however, rather more than half the cases of carcinoma involved its upper part, as in the patient here described.

Of primary malignant neoplasms of the trachea squamous-cell carcinoma is the commonest. Males are affected rather more often than females, by both simple and malignant tumours, in the proportion 3 : 2.

The majority of cases of carcinoma of the trachea show local metastases only, but in 3 recorded cases metastases in the bones of the forearm (Fisher, 1944), widespread metastases in the skin (Keeney, 1947), and metastases in the liver and peripheral lymph-nodes (Zarowitz and Hoffman 1952) are described.

Table I.—SHOWING HISTOLOGICAL TYPES AND NUMBER OF TUMOURS OF TRACHEA REPORTED IN THE PERIODS 1898–1945 AND 1946–51

HISTOLOGICAL TYPE	NUMBER REPORTED					
	1898–1945*		1946–51		Total	
	Benign	*Malignant*	*Benign*	*Malignant*	*Benign*	*Malignant*
Papilloma	67	—	2	—	69	—
Adenomas	19	—	1	—	20	—
Mixed salivary	5	—	1	—	6	—
Cylindroma	1	—	11	—	12	—
Intratracheal goitre	31	—	1	—	32	—
Carcinoma	—	187	—	34	—	221
Lipoma	5	—	—	—	5	—
Fibroma	36	—	2	—	38	—
Chondroma and osteoma	75	—	—	—	75	—
Lymphoma, angioma	9	—	1	—	10	—
Rhabdomyoma	1	—	—	—	1	—
Giant-cell tumour	1	—	—	—	1	—
Sarcoma	—	34	—	2	—	36
Carcinosarcoma	—	1	—	—	—	1
Hæmangio-epithelioma	—	2	—	2	—	4
Endothelioma	—	1	—	—	—	1
Amyloid tumour	3	—	18	—	21	—
Total	253	225	37	38	290	263
Doubtful histology	29					

* After Ellman and Whittaker.

SUMMARY

A patient with carcinoma of the trachea is described whose clinical picture was confused by a co-existing fibroma of the pleura.

The condition is briefly discussed and a tabular summary of the relevant literature is given (*Table I*).

I wish to thank Dr. G. Batten for the radiological report and Dr. Maurice Shaw for permission to publish details of his patient.

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ENDOMETRIOSIS OF THE APPENDIX

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SAMPSON (1922) described the appendix as a frequent site of intestinal implantation of ovarian hæmatoma of endometrial type and found it involved in 4 out of 12 cases. In studies of endometriosis, Smith (1929) found the appendix involved in only 1 case in a series of 200, and Hayden (1942) 1 in a series of 569 cases. Scott (1950) described superficial and small spots on the appendix in 7 out of 516 cases. Collins (1951) reported 9 cases, Sutton and Hardy (1952) 15 cases, and Romanus (1952) 5 cases of endometriosis of the appendix.

CASE REPORT

HISTORY.—M. M., an unmarried woman aged 27 years, was admitted on May 9, 1952, with a history of abdominal pain of 24 hours' duration, which seemed typical of an attack of acute appendicitis. Menstruation had always been regular and her last period was fourteen days ago.

ON EXAMINATION.—She was an obese young woman of good general health and was afebrile. There was guarding and tenderness in the right iliac fossa, being most marked over McBurney's point, and release tenderness was also present. On rectal examination there was a firm, fixed, and tender mass anteriorly and on the right side. A diagnosis of acute appendicitis was made.

AT OPERATION.—The abdomen was opened through a right oblique muscle-splitting incision and appendicectomy performed. The appendix was found to be lying over the pelvic brim with its tip firmly anchored to the deepest portion of the rectovaginal pouch. There was considerable oozing on dissecting it out. Pelvic examination, which was difficult owing to the obesity and the high incision, revealed a normal uterus with a dense mass in the rectovaginal pouch and around the right tube. The right ovary was not palpated and was presumed to be lying in the 'inflammatory mass'. The abdomen was closed in layers.

The patient made an uneventful recovery and was discharged on the tenth post-operative day.

PATHOLOGY.—The appendix measured 60 mm. long and 10 mm. in diameter. Part of the lumen was obliterated. Histologically it showed no evidence of active inflammation. There was some increase of submucosal collagen. Near the tip, within the muscularis, there was a 5-mm. nodule (*Fig.* 408) consisting of smooth muscle in which there were several small endometrial glands, surrounded by a small amount of the characteristic stroma (*Fig.* 409). The nodule resembled one of adenomyosis uteri more than ordinary endometriosis. Near this nodule, but in the subserosa outside the muscularis, there were several small areas of typical endometriosis without associated muscle (*Fig.* 408). There was fresh hæmorrhage around many of the glands, especially the subserosal glands.

FOLLOW-UP.—When seen a month later the patient complained of severe dysmenorrhœa. There were no masses felt in the abdomen or in the operation scar. She was referred to the gynæcology department where she was examined on July 2.

FIG. 408.—Longitudinal section of tip of appendix showing: A, Lumen of appendix; B, Nodule of muscle with endometrial glands; C, Subserosal endometriosis. H. and E. (× 5.)

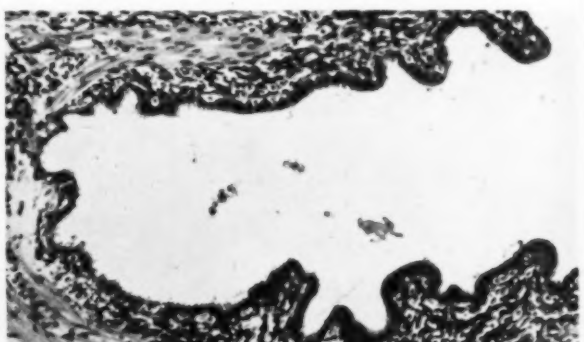

FIG. 409.—A higher power view of the largest gland within the nodule (B) in *Fig.* 408. Showing endometrial type of epithelium and stroma. H. and E. (× 105.)

History now revealed that she had pain after defæcation for the last three years. She also complained of left-sided abdominal pain since June. On examination she was tender in the left iliac fossa. There was a mass felt on the left side on rectal examination. Vaginal examination under anæsthesia revealed a cystic mass in the left fornix.

AT SECOND OPERATION.—The abdomen was opened through a midline subumbilical incision, dense adhesions were found in the pouch of Douglas and a right-sided ovarian cyst was seen displacing pelvic contents to the left. A right oöphorectomy was performed. Convalescence was uneventful and when seen on Dec. 10 she was fit and well, having menstruated normally three weeks previously.

The specimen showed remnant of ovarian tissue with numerous areas of endometriosis, especially on the surface.

DISCUSSION

Sutton and Hardy (1952) in their series of 15 cases found that the 2 cases which were diagnosed as acute appendicitis were the only two in which endometrial tissue was found in the muscularis; the other 13 cases showed only superficial involvement.

Though this patient had endometrial tissue in the muscularis her symptoms were probably due to the recent hæmorrhage under the serosa of the appendix. This, occurring 14 days after the last menstrual period, is unusual in a patient who is regular.

Sampson's (1922) and subsequent findings of right ovarian endometriosis in all cases of endometriosis of the appendix is substantiated by this case. But Deacon (1949) and Collins (1952) describe 2 cases of endometriosis of the appendix without any involvement of the right ovary.

SUMMARY

A case of endometriosis of the appendix is described, which was treated as an acute appendicitis with appendicectomy. Subsequently right ovarian endometriosis was discovered and right oöphorectomy performed with good results.

I wish to thank Mr. R. H. Franklin and Mr. I. Donald, under whose care this patient was admitted, for permission to publish these notes ; Dr. B. Lennox for the pathology reports ; and Mr. E. V. Willmott for the photographs.

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SARCOMA OF THE EPIDIDYMIS

BY T. J. EASON and D. H. K. SOLTAU, LONDON

MALIGNANT tumours of the epididymis appear to be sufficiently rare to be worth recording. A case of sarcoma is reported here and the literature briefly surveyed.

CASE REPORT

A single man, a labourer, aged 43, was admitted to hospital on Jan. 10, 1952, with a right-sided scrotal swelling. He stated that he had noticed a small swelling in this region for two years and that it had become rapidly larger in the last six months. There was no pain, but a feeling of heaviness and discomfort. He did not think he had lost weight, and there was no history of trauma.

Five years ago he had been operated on at another hospital for a right inguinal hernia. His general health had always been good.

ON EXAMINATION.—He was of healthy appearance, with findings as follows :—

Scrotum.—The skin was normal and non-adherent, though the presence of several large veins was noted. A firm, solid swelling, approximately 5 in. by 3 in. and non-translucent, was present on the right side, with a small swelling (subsequently confirmed to be the testicle) at the lower pole. Sensation was markedly diminished, but the cord above the tumour felt normal. The left testicle and cord were normal, and rectal examination was negative.

Abdomen.—Liver and spleen were not enlarged, and no abnormal mass was felt. No lymph-node enlargement. Scar of right herniotomy incision.

Chest.—No abnormal physical signs. X-ray examination negative.

It was decided to explore the scrotal swelling, with the provisional diagnosis of a tumour of the testicle or a chronic hæmatocele.

AT OPERATION.—On Jan. 11 (T. J. E.), under general anæsthesia. Right inguinoscrotal incision. A large tumour with distended veins on its surface was partly delivered from the scrotum when its malignant nature was revealed. The cord was separated and divided as high as possible at the internal inguinal ring, to prevent any dissemination during further manipulations. The tumour was freed from the scrotal wall and removed complete with the testicle and cord. The inguinal canal was repaired and the skin incision closed with a rubber tube drain in the scrotum.

The post-operative course was uneventful, and the wound healed by first intention.

On Feb. 25, after the pathological report of sarcoma had been received, a course of radiotherapy to the abdominal para-aortic and scrotal areas was begun. He was discharged from hospital on Feb. 9, subsequently attending as an out-patient.

Radiotherapy was completed on March 7, a total dosage of 3529 r having been given over twenty-six days.

Follow-up.—

April 1 : He was well, and the scar was soundly healed. He stated that he was returning to Ireland.

FIG. 410.—Photograph of specimen.

May 14 : His doctor wrote to say that he was well, at work, and was putting on weight.

Aug. 31 : A letter was received from the patient in reply to an inquiry stating that he was now working on a farm in Lincolnshire and was keeping very well.

Aug. 21, 1953 : Inquiry elicited letter from patient in Ireland stating that although he felt well he had noticed a fresh swelling recently in the same place. He has been asked to attend personally for review should he return to England.

SPECIMEN.—A solid tumour of fleshy appearance, measuring 4½ in. (11·5 cm.) by 3½ in. (9 cm.) by 2½ in.

(6·5 cm.), arising from and replacing the epididymis, and lying above and behind the right testicle. The tumour did not invade the testicle at all, though it was adherent to it. (*Figs.* 410, 411.)

Histological examination (Dr. L. Hewlett) showed the tumour to be a spindle-celled sarcoma of the epididymis.

FIG. 411.—Photograph showing longitudinal section of specimen, opened out to show tumour, testis, and spermatic cord.

There was no infiltration of some adjacent muscle tissues which had been removed with the tumour. (*Fig.* 412.)

The testicle measured 1 in. by 1¾ in. and was normal on section.

DISCUSSION

Willis (1948) states that epithelial tumours of the epididymis and ductus deferens are very rare, and a survey of the literature reveals very few cases of sarcoma.

O'Brien (1942), reporting a case of primary sarcoma of the epididymis, quotes a personal communication from Gilbert recording 91 cases of malignant tumours, of which 7 were primary fibrosarcomas.

Thompson, of the Mayo Clinic (1936), in an analysis of reported tumours of the epididymis, found 40 per cent were benign and 60 per cent malignant. Of 36 malignant cases 23 were of epithelial origin, 12 were sarcomatous, and 1 a teratoma.

Lazarus (1938) reported 40 cases of malignant disease, 17 of which were listed as sarcomas, with 1 of these classified as a spindle-celled sarcoma. He discussed the possible influence of previous trauma. In 25 cases there was no information, in 10 a positive history of trauma, in 5 a definite absence of trauma.

Charache (1939) combined the cases collected by Lazarus with those of Scalfi (1936). The latter reported 18 benign and 40 malignant cases. Excluding the cases common to both Lazarus and Scalfi, the total number of malignant cases was 54. Of these there was 1 spindle-celled and 1 fibrosarcoma.

Hapert and Thompson (1947) reported a further case of fibrosarcoma of the epididymis.

The case reported by us is unusual in that the size appears to be larger than the majority previously recorded, and the duration of the history (two years, with rapid enlargement in the last six months) longer than most.

Accurate pre-operative diagnosis appears to be very rare, and in this case was not made, the case being considered one of testicular neoplasm. There was no history of trauma.

O'Brien, in his discussion, states that the usual sites of metastatic tumours of the epididymis are the same as in testicular tumours. In most cases lymphatic spread precedes venous. He does not differentiate, however, between carcinoma and sarcoma of the epididymis.

Treatment in this case was conducted on similar lines to that of testicular tumours. Local excision, with high ligation of the cord early in the operation, was followed by high-voltage X-ray therapy to the scrotal region and the para-aortic lymphatic field.

Lazarus also used a combination of surgery and radiotherapy in treatment, and reported 5 out of 40 cases well after 1 year, 1 well after 2½ years, and

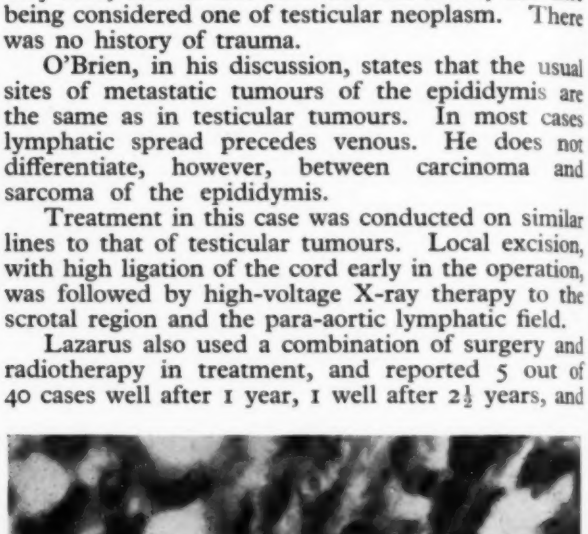

FIG. 412.—Photomicrograph to show histology of the condition. (× 750.)

1 case which died 12½ years later, though the cause is not stated.

However, the prognosis is obviously difficult to assess, as apart from the rarity of these tumours most of the literature deals only with early cases, as here.

SUMMARY

A case of spindle-celled sarcoma of the epididymis is reported. The patient was alive and well 19½ months later although with possible recurrence. The incidence, pathology, and treatment of such tumours are discussed, and the relevant literature reviewed briefly.

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A CASE OF BILATERAL MYCOTIC ANEURYSM OF THE POSTERIOR TIBIAL ARTERY TREATED SUCCESSFULLY BY EXCISION

By G. C. TRESIDDER and R. P. WARREN, London

The case here described comes under the currently accepted definition of mycotic aneurysm—"an aneurysm due to septic infection in the wall of an artery, usually associated with bacterial endocarditis" (Goadby, McSwiney, and Rob, 1949). In this case embolic phenomena were followed a few days later by the formation of a mycotic aneurysm. Mycotic aneurysm is an uncommon complication of subacute bacterial endocarditis, with a poor prognosis. On this occasion the end-result was satisfactory after an exceedingly stormy course.

CASE REPORT

Mrs. R. W., aged 25, was admitted to The London Hospital under Dr. Clark-Kennedy on July 14, 1952. For some weeks before admission she had looked pale and was losing weight. Two weeks before admission she complained of joint pains in the wrists, ankles, knees, and elbows.

On Examination.—A pale, ill-looking woman; temperature 99°, pulse 108. The left ankle was tender and swollen. *Cardiovascular system*: the heart was not enlarged. There was a systolic murmur all over the precordium, and an early diastolic murmur was heard to the left of the sternum. B.P. 130/60. Hæmoglobin 55 per cent.

A provisional diagnosis of acute rheumatic fever and aortic incompetence was made. Salicylates were prescribed but there was no response. On July 21 a blood-culture yielded a growth of *Str. viridans*, sensitive to penicillin and aureomycin. The diagnosis was then changed to that of subacute bacterial endocarditis, and a course of systemic penicillin was started (10 million units a day).

On July 27 she complained of pain in the right forefoot; the toes became discoloured, and the pulsation in the dorsalis pedis artery impalpable. Priscol was prescribed and the circulation gradually improved, except for the tips of the toes, where a line of demarcation developed. The pain gradually subsided as the circulation improved. On Aug. 7 she began to get cramping pains of increasing severity in the outer side of the left leg, and after a few days a similar pain began in the right leg. At this time the early diastolic murmur was more easily heard in the aortic area of the precordium. The pain in the left leg improved but that on the right side became rapidly more and more severe. Although analgesics were prescribed in full and heroic doses, the pain was not controlled. The patient was constantly sobbing and crying out, and was resentful of any attention or examination.

Examination on Aug. 20 showed that on the left side the foot was normal with palpable pulsation in the posterior tibial and dorsalis pedis arteries. In the upper third of the leg, in the peroneal region, there was a tender swelling showing expansile pulsation. On the right side the forefoot was cold, and there was dry gangrene of the terminal phalanx of the second toe and of the tips of the other toes. No pulsations were felt in the posterior tibial or dorsalis pedis arteries. In the calf posteriorly there was a large tense exquisitely tender swelling, with visible and palpable expansile pulsation. On each side compression of the femoral artery stopped the pulsation and relieved the pain. In view of the severe pain it was decided to operate on the right side first. Hæmoglobin 61 per cent.

At Operation (G.C.T.) (Aug. 23).—The right leg was explored through a long posterior incision extending from 4 in. above the knee to just above the medial malleolus. The popliteal artery was isolated above the aneurysm, and the posterior tibial artery below. The gastrocnemius muscle was split vertically and on incising the soleus the aneurysm was opened. The hæmorrhage was controlled by inflation of a pneumatic tourniquet. The friable wall of the aneurysm, which was 5 in. by 3 in., was removed piecemeal. The posterior tibial artery was tied above and below the aneurysm, and the peroneal artery tied below. Faint pulsation could now be felt in the two distal arteries. The wound was closed in layers with drainage down to the site of the aneurysm. Four-and-a-half pints of blood were transfused during operation.

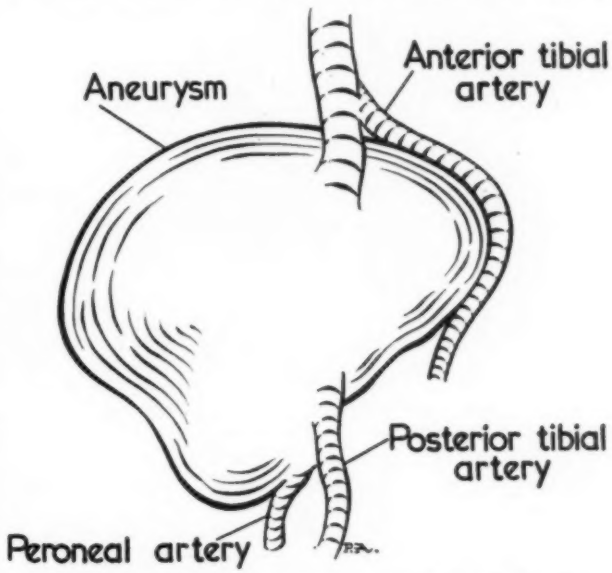

Fig. 413.—Diagram of aneurysm of posterior tibial artery.

Following the operation, the pain in the right leg was very much better and by Aug. 25 pulsations were palpable in the dorsalis pedis artery on the right side and the forefoot was warmer. On this date the pain became more severe in the left leg, the pulsatile swelling became much larger posteriorly, and pulsations in the dorsalis pedis and posterior tibial arteries were no longer palpable. Hæmoglobin 84 per cent.

Second Operation (R. P. W.) (Aug. 26).—As it was thought that the anterior tibial artery might be involved in the aneurysmal process, it was exposed below the swelling before the posterior incision was made. There was no pulsation in the anterior tibial artery at this time. The further steps in the operation were similar to those on the right side. The aneurysm was 6 in. by 3 in. in size and herniating through the interosseous membrane so as to compress the anterior tibial artery, which was intact. (*Figs.* 413, 414.) The wall of the aneurysm was removed piecemeal. The posterior tibial artery was ligated above and below, and the peroneal artery below, the aneurysm. At the conclusion of the operation the anterior tibial artery was pulsating well and there was faint pulsation in the distal end of the posterior tibial

and peroneal arteries. Two pints of blood were transfused during the operation.

Following the operation, the pain was relieved and the patient's condition gradually improved. There was satisfactory healing of the wounds. Hæmoglobin 92 per cent. The patient began increasing activities in bed and then progressed with further exercises until on Oct. 21 she first got up. On Nov. 15 she was first walking with the aid of physiotherapists, and by Dec. 6 was

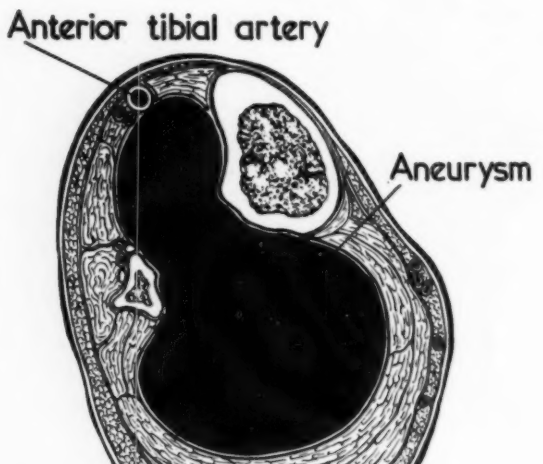

FIG. 414.—Diagram to show compression of anterior tibial artery by aneurysm.

walking unaided. On Jan. 2, 1953, she returned home and was well when later seen in Out-patients.

The patient was treated with systemic penicillin from July 22 to Oct. 13—a total dosage of over 817 million units. She also had a three-weeks' course of aureomycin—total dosage of 42 g.—over the period of the operations.

HISTOLOGICAL REPORT (Dr. D. J. O'Brien).—*Mycotic aneurysm of left posterior tibial artery*: Tissue received consisted of (1) four grey-brown sheets (the largest 5·0 × 2·8 × up to 0·4 cm. thick), one surface smooth the other ragged; flakes of friable grey fibrin could be detached from the surfaces; (2) a plaque (2·5 × 2·0 × 1·0 cm. thick), the inner surface partly smooth, partly wrinkled, two vessels projected from the fibrous outer surface.

MICROSCOPICAL.—The sheets of tissue from the aneurysm wall are composed of a narrow layer of collagen varying slightly in thickness, with, on the inner surface, laminated fibrin thrombus in which there is no excess of leucocytes. Outside the collagen layer is a broad zone of pyogenic granulation tissue with focal areas of necrosis.

The elastic coats of the arterial wall are not present in the collagen layer, but in the outer zone are the ghosts of necrosed vasa vasorum. Gram-positive and -negative cocci—singly and in small clumps and chains—are present in the outer zone.

There is no inflammation of the adventitia or media of the arteries at their entry into the aneurysm. The wall remains intact for a very short distance only, the media rapidly narrows, and the elastic layers fragment and disappear. At the junction of artery and aneurysm there is in the place of the intima a band, equal to the media in thickness, of loosely and irregularly arranged spindle cells in a pale hæmatoxyphil matrix. This band is not infiltrated with leucocytes.

Farther away from the mouth of the artery collagen fibrils increase in this band, and it continues beyond the end of the media to become the central collagen layer of the aneurysm wall.

It is not clear whether this tissue is derived from proliferation of the intima, or is an ingrowth from the outer part of the aneurysm, but the absence of capillaries favours an intimal origin.

BACTERIOLOGICAL REPORT.—Swabs taken from the wall of the aneurysm were sterile on culture.

COMMENT

An unusual feature in this case was the symmetrical and almost simultaneous appearance of the lesions, which were situated on the posterior tibial arteries just after the peroneal branches had been given off. As far as can be determined from the literature no similar bilateral lesion with recovery following operation has previously been recorded. It is interesting to note that the penicillin was not effective in preventing the formation of the aneurysms. Nor, presumably, was the infection of the vegetations controlled immediately.

The aneurysms appear to have been present for about three weeks before operation, which is too short a time for collateral vessels to develop. The maintenance of an adequate circulation following operation, in which the aneurysmal part of the posterior tibial artery was resected, seems to be due to the relief of pressure from the aneurysm on neighbouring vessels. This was particularly well shown in the case of the anterior tibial artery on the left side. The other possibility is that the removal of the aneurysm may at the same time have removed the stimulus for sympathetic vasoconstriction, and in this way allowed the return of the normal circulation and thus preservation of the limb.

The main benefit of operation, from the patient's point of view, was the dramatic relief of the pain.

Addendum (September 1953).—The patient is extremely well and has gained 15 lb. in weight since January. The small patches of gangrene have separated from the tips of the right toes. The right dorsalis pedis artery pulsates strongly, but the left is impalpable. There is a valgus deformity of the left foot associated with some pes cavus. Both feet are somewhat œdematous, but this is easily controlled by the use of elastic stockings.

We wish to express out thanks to Dr. Clark-Kennedy for his kindness in referring this patient and for supervising the medical side of her treatment while she was in the surgical ward. Our thanks are also due to Dr. D. J. O'Brien for the pathological report; to Professor Dorothy Russell for her advice; and to Miss P. Archer for the drawings.

REFERENCE

GOADBY, H. K., MCSWINEY, R. R., and ROB, C. Q. (1949), *St. Thomas's Hosp. Rep.*, **44**.

REVIEWS AND NOTICES OF BOOKS

Operative Surgery. By GUY H. HORSLEY, M.D., F.A.C.S., Associate Professor of Surgery, Medical College of Virginia, and ISAAC A. BIGGER, M.D., F.A.C.S., Professor of Surgery, Medical College of Virginia. 2 Vols. Sixth Edition. 10¼ × 7 in. Pp. 1579 + x. 1274 illustrations. 1953. London: Henry Kimpton. £11 5s.

THIS large two-volume work on operative surgery with its numerous contributors covers this ever-growing field in a comprehensive way. Perhaps too much has been incorporated about operative procedures now discarded. Operations which are never used in England to-day are described, this applies especially to amputations.

The special branches of surgery, such as plastic surgery, thoracic surgery, and gynæcological surgery, have been allotted too much space in comparison with that allowed for general surgery, which is a pity.

The illustrations although numerous are not up to the standard we are accustomed to find in American text-books, and they lack definition and there is too little contrast of the tissues in the pictures.

This is an American work which will not be of value to the postgraduate in this country who wishes to obtain higher surgical qualifications, for it does not contain the essentials on which he would be examined over here. One notices that practically all the references to the literature are from American sources.

Fractures of the ribs always unite, and yet in this work three different ways of wiring or pegging fractured ribs are described and illustrated; surely this is unnecessary and useless!

If we have been critical in reviewing this book we hope that the criticism has been constructive.

Pictorial Introduction to Neurological Surgery. By G. F. ROWBOTHAM and D. P. HAMMERSLEY. 7¼ × 9¾ in. Pp. 108 + viii. 1953. Edinburgh: E. & S. Livingstone Ltd. 21s.

IT is refreshing to find a book of this type dedicated to "Charles Macmillan, to whom medical publishing and many authors owe so much". Surgeons both at home and abroad will agree with this statement.

Neurosurgery is a difficult branch of surgery; it is also one which has progressed by leaps and bounds during the last twenty years. This pictorial volume with so many excellent drawings of various operative procedures should prove of great value to all those who wish to acquire knowledge of neurosurgical techniques.

It is essentially a book for postgraduates and it will fill a definite gap in the list of essential books which should be read for the final fellowship examination.

An excellent production and one of which the house of Livingstone may well be proud.

Surgery of Repair as applied to Hand Injuries. By B. K. RANK, M.S., F.R.C.S., F.R.A.C.S., Honorary Plastic Surgeon, Royal Melbourne Hospital; etc.; and A. R. WAKEFIELD, M.S., F.R.C.S., F.R.A.C.S., Honorary Plastic Surgeon, Children's Hospital, Melbourne; etc. With a Foreword by SIR GORDON GORDON-TAYLOR. 6¾ × 9¾ in. Pp. 256 + xiv, with 188 illustrations. Edinburgh: E. & S. Livingtone. 40s.

IN this traumatic age it is essential that surgeons should know how to apply the surgery of repair. In no other field of surgery is this more important than in hand injuries. It is a significant fact that during 1949 in Great Britain, of approximately 737,500 injury benefits for accidents and prescribed diseases during employment, 211,280 were paid to persons with hand injuries. These figures speak for themselves and reveal the great importance not only in the prevention of these accidents but also in the treatment of the damaged hands.

Rank and Wakefield are plastic surgeons with an extensive clinical background both in England and Australia, and they have drawn on this practical experience in writing this book. They have written a very interesting and important volume which is well illustrated and well documented.

We agree with Sir Gordon Gordon-Taylor, who has written a foreword to the book, in that "those whose function it is to deal with injuries of the hand will derive much profit and valuable assistance from those parts of the book drawing attention to what might be termed 'a new anatomy of the hand'; it is of interest to the scholar as well as the surgeon".

An excellent production.

British Medical Science and Practice. An Anthology. By G. F. PETRIE, M.D. London: Longmans Green & Co. 15s.

THIS is an interesting and rather unique anthology which pictures modern trends in British medicine. Part I deals with ten pioneers in British medicine and gives excellent pictures of William Harvey, John Hunter, Jenner, Bell, Bright, Addison, Simpson, Florence Nightingale, Lister, and Sir Patrick Manson. Part II consists of selected aphorisms gathered from sources ranging from the seventeenth to the twentieth century.

Part III is devoted to an anthology of modern British medicine, and extracts from articles written by well-known scientists, physicians, and surgeons are given. These extracts have been very well chosen and make fascinating reading.

An excellent bedtime book for all medical men.

Physical Examination of the Surgical Patient. By J. ENGLEBERT DUNPHY, M.D., F.A.C.S., Assistant Clinical Professor of Surgery, Harvard Medical School; etc.; and THOMAS W. BOTSFORD, M.D., F.A.C.S., Clinical Associate in Surgery, Harvard Medical School; etc. 6⅛ × 9¼ in. Pp. 326 + xiv, with 188 illustrations. 1953. Philadelphia and London: W. B. Saunders Co. 37s. 6d.

THIS book is a product of the Harvard Medical School and reflects the tradition of the School in its presentation. The authors describe the purpose of the book as being "to discover the presence and extent of injury or disease"; the tools to be used are "eyes, ears, fingers, nose and brain"; and stress the fact "that in order to discover the abnormal one must learn and know the normal". They also quote in the preface the invaluable and eminently practical maxim of Dr. William Thayer: "learn to see, hear, feel and smell and know that by practice alone you can become expert". No teacher of clinical surgery will disagree with these details.

One of the most important periods of a medical student's training is the first three months, during which he is passing from his preclinical to the clinical studies.

He has to be taught and trained to apply and correlate his preclinical knowledge to the problems of disordered anatomy and physiology with which he is brought into contact. His powers of observation and synthesis must be developed, and if the methods adopted by the teachers of the three main subjects be co-ordinated and supervised by a good teacher of pathology, then as he progresses

in his studies of clinical medicine all the new subjects he meets will be seen by him in a proper perspective and will finally be co-ordinated in his mind as a composite whole.

The book consists of a series of chapters on clinical methods of examination of the various systems and is divided into two parts, the first being what is called " the elective examination " and the other " the emergency examination ".

The emergency examination deals with injuries which have occurred, usually from severe trauma, in which it is urgently necessary that a proper assessment of the extent of the injury should be made as soon as possible. The elective examination is that which can be made more leisurely but no less accurately, and in its discussion will open up questions involving differential diagnosis, pathology, and treatment, but these latter aspects are not unduly stressed.

It is a well-balanced book and conveys to the student just those essentials of clinical medicine which he must master early in his training. It is not what so many books become, when dealing with the same subject, a hotchpot of all aspects of medicine, surgery, gynæcology, and pathology, together with all the ancillary subjects in their various ramifications, with no definite co-ordination or stress on the most important.

The line drawings with which the book is illustrated should be so much more convincing to the student than the photographs which are now used so often to explain the text, e.g., there is only one radiograph in the book, and no actual photograph of a case.

There is a valuable chapter on the method of examination of the abdomen in infants and children, and another on the pitfalls of abdominal examination, both of which are as a rule insufficiently stressed to the junior student.

The chapter on peripheral vascular diseases and gangrene are what one would expect from the school of Dr. John Homans, and the figures taken from his text-book are as convincing and explanatory as on the first day they were published.

In some schools the preliminary course introducing the students to clinical studies is called ' shepherding '. It would be wise if the shepherd introduced this book to the sheep and followed its precepts.

The Faber Medical Dictionary. Edited by Sir CECIL WAKELEY, Bt., K.B.E., C.B., LL.D., President of the Royal College of Surgeons of England. $7\frac{1}{4} \times 10\frac{1}{2}$ in. Pp. 471 + vii. 1953. London: Faber & Faber. 45s.

IT is refreshing to see a new medical dictionary of English origin, for there never was a time when such a book was more in demand.

Sir Cecil Wakeley has edited a concise, modern, up-to-date dictionary which will prove of real value not only to doctors, nurses, and dental surgeons, but to countless others in the many and varied medical auxiliaries in the Health Service. The great and important organizations, such as the Red Cross and the Order of St. John, with its vast ambulance brigade, all need a dictionary of this type, for in it they will find a brief explanation of the medical and surgical terms which frequently confront them. There can be no doubt that such a book as this is a much-needed source of reference for hospital administrators, medico-social workers, hospital secretaries, and the vast field of people who are in some way connected with medical work.

Sir Cecil Wakeley has certainly done a fine job, but we all expect this of him, for he has edited many books and journals for many years. This new medical dictionary is surely one of his best efforts and the medical profession as a whole is greatly indebted to him for this book.

An excellent production.

Basic Pathology and Morbid Histology. By D. B. CATER, M.A., M.D.(Cantab), F.R.C.S.(Eng.), University Demonstrator in Pathology, University of Cambridge. $6\frac{1}{2} \times 9\frac{1}{4}$ in. Pp. 330 + viii, with 264 illustrations, including 20 fully coloured plates. 1953. Bristol: John Wright & Sons Ltd. 42s.

THIS is a novel type of book, for it embraces basic pathology, a subject which is not dealt with in the bigger text-books of pathology. The modern student will welcome this book because it presents him with exactly what he wants. Pathology is a difficult subject and one which causes the average student much concern because of its complexity and mass of detail. Dr. Cater has produced an excellent book, full of elementary factual information, told in a simple style, and, what matters most, in an interesting manner.

One of the features of this book is the excellence and originality of the illustrations. The coloured illustrations are superb and could not be bettered. Good illustrations are said to be worth a thousand pages of text, and this is certainly true of this book. Students at home and abroad will consider this book as essential to their own personal library. Our only regret is that the book was not available when the reviewer was a student, for it would have been of real value to him.

Surgical Pathology. By LAUREN V. ACKERMAN, M.D., Professor of Surgical Pathology, Washington University School of Medicine. Pp. 836, with 913 illustrations. 1953. London: Henry Kimpton. £5 10s.

ALTHOUGH the author states that " this book can only be an introduction to the vast field of surgical pathology ", yet one must admit he has covered his subject in a very comprehensive manner. Surgical pathology increases year by year and is to-day a vast subject. The reaction of the tissues of the body and tumours to X-ray therapy, radio-active isotopes, hormones, and cortisone make a study in themselves.

Professor Ackerman, with his matured clinical background acquired during his extensive studies in pathology, gives us a well-balanced book in which it is easy to find any subject which could be said to come under the umbrella of surgical pathology. The illustrations are excellent and include clinical cases, macroscopical photographs, and photo-micrographs. This is the real way to understand surgical pathology, and students and postgraduates alike will find this book of real value.

We can agree with the author in his distrust of grading of breast cancer and the ultimate prognosis. There is poor correlation between grading and metastases.

The surgeon will find much of interest in this large volume and as a reference book it is practical and up to date.

An Introduction to Abdominal Diagnosis. A Handbook for Students and Practitioners. By ALAN E. LEE, M.D. (Melb.), F.R.C.S. (Eng.), F.R.A.C.S., Senior Visiting Surgeon, Brisbane Hospital, Brisbane. $5\frac{7}{8} \times 9\frac{1}{8}$ in. Pp. 176. 1951. Sydney: Australasian Medical Publishing Co. Ltd.

IT is right that a student entering the surgical wards should try to apply his knowledge of physiology to the interpretation of symptoms. This book shows him the lines on which to think and to examine his patients, and the second chapter in particular gives a good, if somewhat theoretical, account of the physiological basis of pain.

It will also give the surgeon and practitioner a guide to diagnosis which will help to elucidate his difficult cases.

The book is clearly written and most readable; the type is of good size and the paper excellent.